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Case Report

Tubercular pyometra in a young unmarried female - dilemma and pitfalls in diagnosis: a rare case report with review of literature

Rasika Aggarwal, Renuka Malik*, Swati Singh

Department of Obstetrics and Gynecology, ABVIMS, New Delhi, India

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***Correspondence:**
Dr. Renuka Malik,
E-mail: renucam@yahoo.co.in

ABSTRACT

This rare case is the first case being reported as tubercular pyometra in a young unmarried woman. Diagnosis of genital tuberculosis which is a form of EPTB (extra pulmonary TB) can be baffling, compelling a high index of suspicion owing to paucibacillary load in the biological specimens. A negative smear for acid-fast bacilli, lack of granuloma on histopathology and failure to culture mycobacterium tuberculosis do not exclude the diagnosis of EPTB. A 25-year-old unmarried, government employee from Bihar presented to our OPD with secondary amenorrhea for two months carrying with her an USG, CT and MRI done in Bihar suggesting enlarged uterus with fluid collection. CT-also reported few enlarged lymph nodes. Her preoperative investigations revealed an elevated ESR and x-ray chest was normal. Dilatation was done under ultrasonic guidance in OT and 150 cc of thick caseous material was drained A gentle curettage was done on lateral wall near cornea and both the caseous material and endometrial tissue was sent for gram staining, TB-PCR (polymerase chain reaction), NAAT (nucleic acid amplification techniques) and culture. In the post-operative period gram staining for AFB, NAAT, TB-PCR all came negative and it was difficult to convince patient to take ATT. However, on day 10, HPE report came as granuloma suggestive of TB and patient was put on ATT. Culture too was reported negative later. Paucibacillary female genital TB (FGTB) is difficult to diagnose because of varied presentation and limitations of diagnostic tests A raised ESR is presumptive but non-specific. Other tests are x-ray chest, HSG, endometrial tissue for TB PCR nucleic acid amplification techniques (NAAT, HPE and culture (conventional or Bactec). Patients with EPTB are, however, more likely to have negative sputum smear results and many EPTB cases do not have direct lung involvement. Currently, there are no standard guidelines or algorithm for the diagnosis of FGTB. Female genital TB has varying presentation and diagnosis is difficult because of the paucibacillary nature.

Keywords: Genital tuberculosis, Pyometra, Secondary amenorrhea

INTRODUCTION

Genital tuberculosis is a form of extra pulmonary tuberculosis which is usually secondary to pulmonary Kochs, meningeal or bony tuberculosis. The fallopian tubes are involved in 90-100% cases, endometrium is involved in 50-80% cases, ovaries are involved in 20-30% cases, and cervix is involved in 5-15% cases of genital TB. Tuberculosis of vagina and vulva is rare (1-2%).[1] The mode of infection can be commonly haematogenous or less common thru direct, lymphatic spread or even through sex.[2] The patient can present with amenorrhea, oligomenorrhea, pain abdomen, infertility, tubo-ovarian masses, Asherman syndrome as pyometra in menopausal women or as emergency as tubercular peritonitis or as spontaneous rupture of tubercular pyometra.

A rare case of a tubercular pyometra in a young unmarried girl is being reported for its rarity and

difficulty in positive tests despite drainage of caseous material. Confirmatory diagnosis of pauci-bacillary EPTB poses a diagnostic challenge.

CASE REPORT

Miss X, 26-year-old unmarried female, post graduate, government employee resident of Bihar, upper middle class presented to gynae OPD of RML Hospital, New Delhi on 22th October 2019 with complaints of pain in lower abdomen for 2 years, hypomenorrhea and discharge per vaginum for 8 months. Patient had come from Bihar to the hospital for treatment. Carrying an USG report of large uterine collection done in Bihar. Pain had become progressively severe and associated with vomiting over the last 6 months. She reported decreased appetite and evening malaise since last 1 year. There was no history of urinary or bowel complaints. Her general condition was fair and she was afebrile. Abdomen was soft with no guarding or tenderness. ESR was raised to 108. Her serum TSH and x-ray chest were normal. Urine pregnancy test was negative. Imaging studies (USG and MRI) revealed thick collection in endometrial cavity suggestive of hydro, hemato or pyometra/L ovaries were normal (Figure 1, 2). CT scan of abdomen revealed few enlarged retroperitoneal lymph nodes. A provisional diagnosis of hematometra was made and patient was scheduled for EUA and drainages of hematometra. A diagnostic laparoscopy to see extent of tuberculosis in abdomen and tubes and future possibility of infertility was discussed with the patient and her parents. The patient refused consent for laparoscopy. Patient was taken for ultrasound guided dilatation and aspiration of endometrial contents. Misoprostol 400 mg was put 2 hours prior to evacuation. Serial dilatation of cervix was done till Hegar's no. 8 under ultrasound guidance. The dilatation was smooth with no suspicion of cervical stenosis. On dilatation to size 8 thick white cheesy material came out and Karmen suction cannula was used to empty the cheesy content [Figure 3 (a, b, c)]. About 100-150 cc thick caseous material was aspirated from endometrial cavity. A guided gentle blunt check curettage was done.

Figure 1: Ultrasound of collection in endometrial cavity.

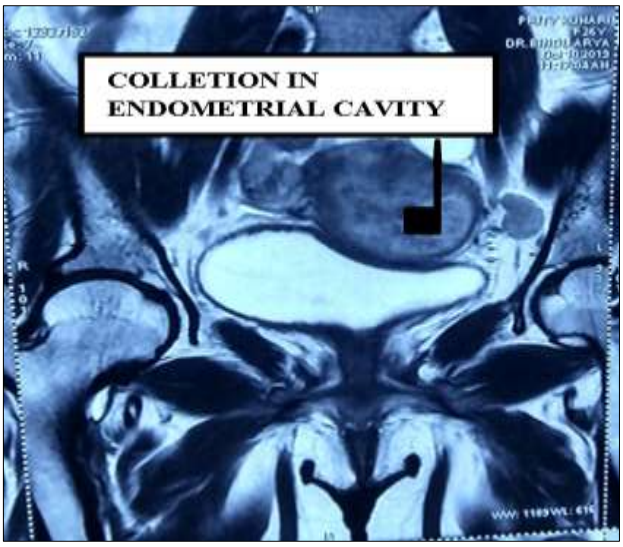

Figure 2: MRI of collection in endometrial cavity.

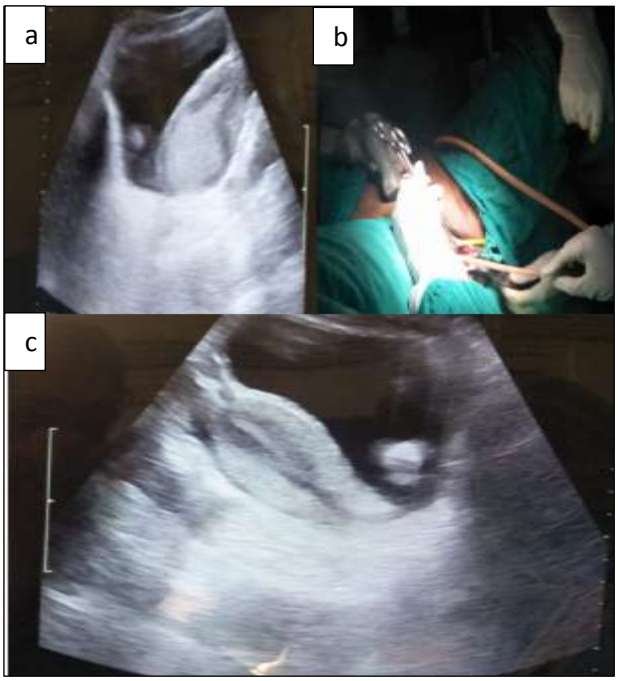

Figure 3: (a) Intraoperative USG of endometrial; (b) USG guided aspiration being done; (c) Postoperative ultrasound of empty endometrial cavity.

The caseous material was sent for AFB, (acid fast bacilli) stain, culture and sensitivity, TB-PCR and CB-NAAT and histopathological examination. Post procedure USG showed empty uterine cavity. Patient was informed of the findings and the reports were awaited. AFB stain, and CB-NAAT turned out to be negative for TB. On day 2, The patient was put on antitubercular therapy in view of caseous material aspirated suggestive of tuberculosis while waiting for PCR, histopathology and culture reports. As per Government of India guidelines the disease was notified. After one day of treatment patient refused ATT because of nausea and vomiting and wanted

positive confirmation of TB. On day 3, PCR report turned out to be negative for TB. A repeat endometrial aspiration and sample was sent for TB-PCR on day 8 hoping to convince the patient to take ATT. The repeat specimen also came out to be negative. However histopathological examination revealed sub epithelial epitheloid cell granuloma along with giant cells suggestive of tuberculosis (Figure 4 (a) and (b)]. ATT was restarted as patient was now convinced seeing a positive report. She was put on daily therapy of Rifampicin, Isoniazid, Pyrazinamide and Ethambutol for 2 months followed by daily 4-month therapy of Rifampicin and Ionized. Patient left for her hometown in Bihar. A follow up, 2 months later telephonically post treatment revealed that she is still ammenoric and she was told to take estrogen for 3 months followed by progesterone for14 days.

In the 5-month follow-up period patient has remained amenorrheic after E+P. She had complained of severe lumbar pain. Subsequent MRI Spine showed multiple psoas abscess in thoraco-lumbar area.

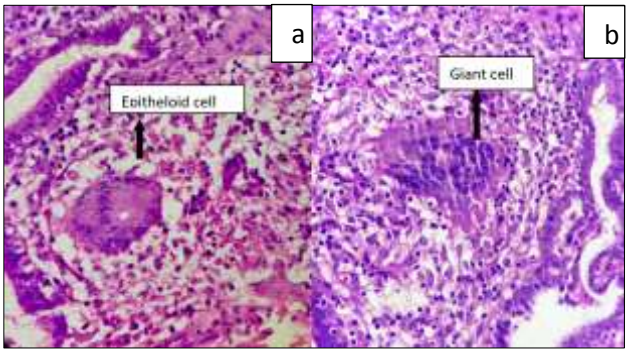

Figure 4: (a) Histopathology of endometrium showing epitheloid cell granuloma (b)Histopathology of endometrium showing giant cell.

DISCUSSION

Tubercular pyometra in a young female is very rare. Using web search engine Google, Pub med and Medline, only one case of tubercular pyometra in a young married female with infertility was found from India.[3] To the best of the knowledge this case of pyometra in a young unmarried girl is the first case reported so far. Most cases of tubercular pyometra are seen in post-menopausal women having senile atrophic cervix causing stenosis.[4] A rare case was reported from morocco by Lahbabi M as tuberculosis peritonitis in pregnancy.[5]

Genitourinary TB is a common form of extra pulmonary TB (EPTB) Worldwide (27%) with genital TB alone accounting for 9 per cent of all EPTB cases. The presentation can be varying like lower abdominal pain, infertility, scanty periods, secondary amenorrhea, post-menopausal bleeding. However, the burden of genital TB in females is underestimated as most of the patients are asymptomatic and usually diagnosed during evaluation for infertility. A study among women with infertility registered for in vitro fertilization in north India reported the prevalence of genital TB in patients with tubal factor infertility as 48.5 per cent.[6]

In female genital TB, the fallopian tubes are involved in 90-100% cases, endometrium is involved in 50-80% cases, ovaries are involved in 20-30% cases and cervix is involved in 5-15% cases of genital TB. Tuberculosis of vagina and vulva is rare (1-2%).[7] WHO Global report of 2019 reports TB as a major global public health, problem with India having the largest burden There is association of human immunodeficiency virus (HIV) co-infection.[8]

As per the WHO, diagnosis of extra pulmonary tuberculosis (EPTB) should be made on the basis of culture positive specimen, or positive histology or strong clinical evidence consistent with active EPTB.[9]

Due to paucibacillary nature of female genital TB, diagnosis is often difficult as in this case. A raised ESR is presumptive and a positive Monteux test has limited utility in populations with high TB burden and where Bacille Calmette–Guérin (BCG) vaccination is followed as a routine. Other tests are x-ray chest, HSG, TB PCR, nucleic acid amplification techniques (NAAT), HPE and culture (conventional or Bactec) patients with EPTB are, however, more likely to have negative sputum smear results and many EPTB cases do not have direct lung involvement.[10,11] For ZN staining to yield a positive result, a sample should contain 10-10 bacilli/ml. Culture for Mycobacterium is more sensitive and requires 10-100 bacilli/ml of tissue/fluid sample for the diagnostic yield. Liquid cultures require at least 9-10 days for positive results and six weeks for being considered negative and in LJ medium cultures, the minimum time-to-positivity is 4-8 weeks.[2] Currently, there are no standard guidelines or algorithm for the diagnosis of FGTB. The DNA-PCR is unable to differentiate viable and nonviable organisms, while bacterial mRNA positive is would indicate the presence of viable organisms. The mRNA-based reverse transcriptase-PCR (RT-PCR) is a rapid method to differentiate viable and nonviable M. tuberculosis and has also been used for the diagnosis of EPTB as well as to monitor drug resistance; the reason for widely used IS6110 in PCR tests is the presence of its multiple copies in M. tuberculosis complex genome, which is believed to confer higher sensitivity However, a few studies from different geographical regions of the world have reported that some clinical isolates have either a single copy or no copy of IS6110 that leads to false-negative results.[12,13]

In the past decade, nucleic acid amplification techniques (NAAT) have been used which have high positive predictive value (98-99%) and relatively lower negative predictive value for early detection of M. tuberculosis from various extra-pulmonary clinical specimens.

The advantages of amplification technique over culture are higher sensitivity, as it can detect as few as 1-10 organisms and are quick to give result in 6-8 hours. NAAT has been

recommended by WHO for diagnosis of TB in 2011. However, in case it had come negative for TB.

Since the patient was unmarried, a diagnostic laparoscopy to see extent was refused by the patient and HSG was not indicated. In the patients presenting with infertility hysterosalpingogram may show distorted endometrial cavity, beaded appearance of the tubes, retort shaped hydrosalpinx, calcified areas and cornual blocks are seen. On laparoscopy, tubercles on peritoneum, tubo ovarian masses caseous nodules, encysted ascites, pelvic adhesions hydrosalpinx, pyosalpinx beaded tubes tobacco pouch appearance may be seen and sometimes it may not be possible to visualize tubes due to adhesions and biopsy for HPE and culture may be taken.[13,14] The CT Whole abdomen was reported as few enlarged lymph nodes that could be suggestive of primary abdominal tuberculosis.

To prevent over diagnosis and to tackle emerging resistance to ATT government of India has made TB as a notifiable disease in 2019. However, the need of a highly sensitive, good specificity and rapid test is still unmet and more research is needed on developing a good lab test.

CONCLUSION

The diagnosis of paucibacillary genital tuberculosis remains challenging and a variety of tests along with clinical findings are needed for diagnosis.

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