Case Report

Pregnancy through Assisted Reproductive Technology in a Patient with Thoracic Endometriosis Syndrome

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Abstract

Extrapelvic endometriosis is known to be difficult to diagnose. Appropriate management in an infertile patient with extrapelvic endometriosis is not only difficult but also not well established. This case report describes a patient with thoracic endometriosis syndrome who was managed successfully for controlling her pleural endometriosis and achieving a pregnancy through assisted reproductive technology (ART).

Keywords: *Endometriosis, pleurodesis, thoracic endometriosis syndrome, video-assisted thoracoscopy*

Introduction

Endometriosis characterized by the presence of functional endometrial tissue outside the uterus affects 5%–15% of women during their reproductive years.[1] Although usually confined to the pelvis, endometriosis is also known to occur in the extrapelvic organs or tissues; ectopic endometrium has been found in the umbilicus, abdominal scars, breasts, extremities, pleural cavity, and lung.[2-4] Although the symptomatology of extrapelvic endometriosis is always coordinated to the menstrual cycle, this is not directly apparent in all patients and diagnosis is notoriously difficult.[5]

Here, we present a case of a patient who presented with subfertility and during workup was diagnosed to have thoracic endometriosis syndrome (TES), which was managed successfully with *in vitro* fertilization-embryo transfer (IVF-ET), resulting in pregnancy and delivery.

Case Report

A 29-year-old female married for 4 years, presented with subfertility, with no menstrual complaints. All the basic investigations of the couple were normal, including folliculometry. She never gave a history suggestive of endometriosis or other pelvic pathology. Hence, she was advised for a diagnostic hysterolaparoscopy in November 2014. During the routine preoperative workup, an X-ray chest done in the immediate postmenstrual phase showed right-sided hydropneumothorax [Figure 1]. Hence, the surgery was postponed and reference was made to a chest physician, and on his advice, a computed tomography (CT) chest was done. The same revealed moderate hydropneumothorax with passive consolidation in the right upper and middle lobe with mild shift of mediastinum to the left [Figure 2]. The patient had occasional dry cough, but there was no history of fever, breathlessness, or weight loss, excepting right-sided chest and shoulder pain. A provisional diagnosis of Koch's was made by the treating chest physician, and the patient was started on anti-Koch's treatment. The patient was started on anti Koch's treatment on November 25, 2014, empirically. However, within 2 weeks of the treatment, the patient developed side effects of the AKT, and her liver enzymes were deranged. The patient discontinued the treatment and was referred for a second opinion for the same. She was subjected to bronchoscopy which revealed a mildly inflamed right upper lobe bronchus. The bronchial and alveolar lavage was negative for tuberculosis mycobacteria growth indicator tube and also negative for culture. Pleural tapping fluid as also subjected to the same test which also was negative. Adenosine deaminase of the pleural fluid was also negative. Both the fluid was also sent for GeneXpert, which was also not informative. A repeat CT (December 20, 2014) chest

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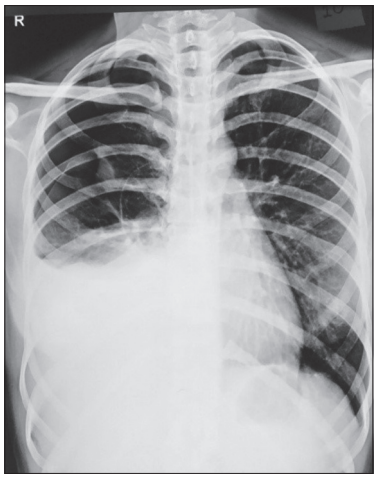
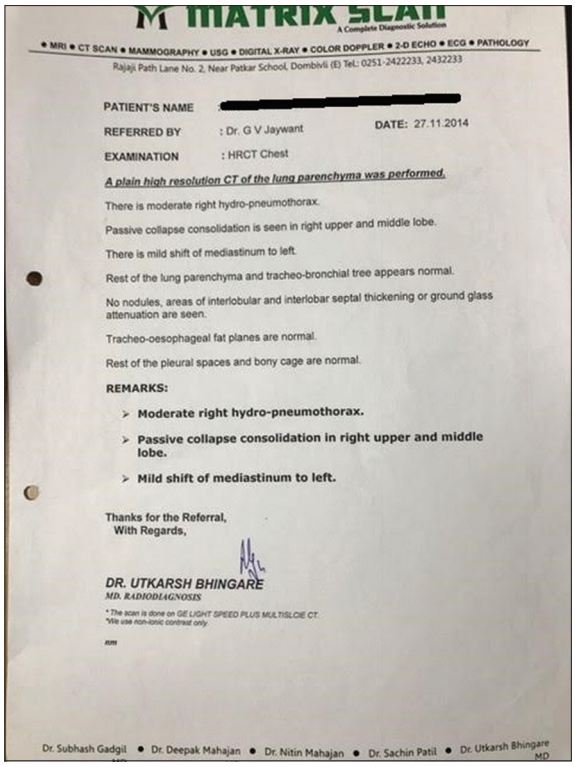


Figure 1: Right hydropneumothorax

revealed the same findings, and hence, an intracostal drainage was inserted [Figure 3]. When the drain was *in situ*, the patient had menstruation with complaints of chest pain in the right side of the chest and the drain tube had more blood, which clinched the diagnosis of "catamenial pneumothorax." The patient was subjected to video-assisted thoracoscopy (VATS) with bronchoscopy on January 28, 2015, in which ectopic endometrial tissues were seen over pleura, lung, as well as parts of diaphragm, were subjected to biopsy and sent for histopathology and culture, and were cauterized. Pneumatocele was identified on the right lobe, for which staples were applied. The histopathology confirmed pleural endometriosis. The tissue was subjected to immunohistochemistry which confirmed the tissue to be positive for estrogen receptor, progesterone receptor, CD-10 – consistent with pleural endometriosis [Figures 4 and 5]. As per the patient's desire, she was subjected to pleurodesis with steritalc and was put on injection leuprolide for 6 months. This gave her good relief from right-sided chest pain with amenorrhea for 9 months. The patient was monitored with a series of chest X-rays and CT scans. Serum CA-125 was not performed in this patient.

The patient returned for infertility treatment in April 2016. Hysteroscopy with uterine septum resection (1–1.5 cm fundal septum – cut with scissors) was carried out on May 26, 2016. Laparoscopy was avoided as the patient had persistent right pleural effusion [Figure 6] (decreased than before). After detailed counseling, including all options, the couple wished to undergo self-stimulation and transfer. Patient's anti-Müllerian hormone was 1.65. The patient was started on IVF antagonist protocol in July 2016 with recombinant follicle-stimulating hormone 225 i.u. daily from day 2 of menstrual cycle. Ovum pick up was done on July 25, 2016, and seven oocytes were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PATIENT'S NAME [redacted]
REFERRED BY : Dr. G V Jaywant DATE: 27.11.2014
EXAMINATION : HRCT Chest

A plain high resolution CT of the lung parenchyma was performed.

There is moderate right hydro-pneumothorax.
Passive collapse consolidation is seen in right upper and middle lobe.
There is mild shift of mediastinum to left.
Rest of the lung parenchyma and tracheo-bronchial tree appears normal.
No nodules, areas of interlobular and interlobar septal thickening or ground glass attenuation are seen.
Tracheo-oesophageal fat planes are normal.
Rest of the pleural spaces and bony cage are normal.

REMARKS:
- Moderate right hydro-pneumothorax.
- Passive collapse consolidation in right upper and middle lobe.
- Mild shift of mediastinum to left.

Thanks for the Referral,
With Regards,

DR. UTKARSH BHINGARE
MD. RADIODIAGNOSIS
* The scan is done on GE LIGHT SPEED PLUS MULTISLCIE CT.
*We use non-ionic contrast only.

Dr. Subhash Gadgil • Dr. Deepak Mahajan • Dr. Nitin Mahajan • Dr. Sachin Patil • Dr. Utkarsh Bhingare MD

Figure 2: Computed tomography chest

retrieved. Six embryos obtained on day 3 (3–8 cell A, 2–8 cell B, 1–8 cell C) and the embryos were electively cryopreserved. The patient received leuprolide depot for one cycle, and frozen embryo was planned in the next month with estradiol valerate 6 mg/day, from the 3rd day of cycle. After 12 days of estradiol valerate, once the endometrium was satisfactory, progesterone support was given and three embryos (2 Grade A, 1 Gr B) were transferred on August 24, 2016.

The patient conceived in the same cycle and was followed up closely throughout the 9 months of pregnancy, which was uneventful. At 36 weeks of gestation, the patient presented with acute urinary retention and elective cesarean section was done on April 19, 2017, and a male child weighing 2.4 kg was delivered. Uterine surface had endometriotic blebs with increased vascularity [Figure 7]. The patient was discharged on day 6 of delivery. Lactation was also well established and has been encouraged to continue breastfeeding till at least 1-year postpartum.

An X-ray chest was taken 3 weeks postpartum [Figure 8].

Discussion

TES is a challenging clinical entity. A high index of clinical suspicion is of paramount importance as both diagnosis and treatment may often be delayed for years. The present case had an important feature that the patient never gave any history suggestive of pelvic

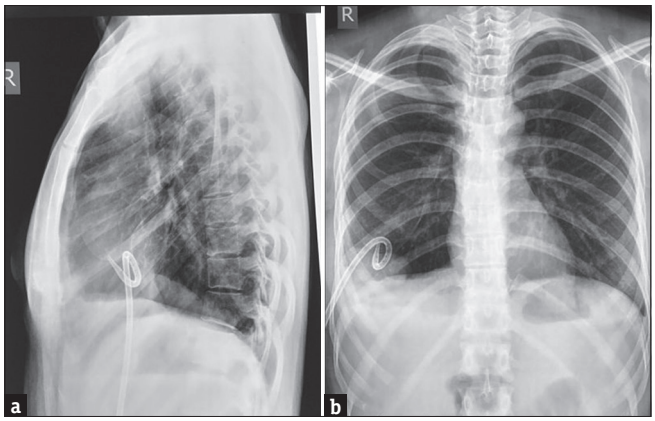
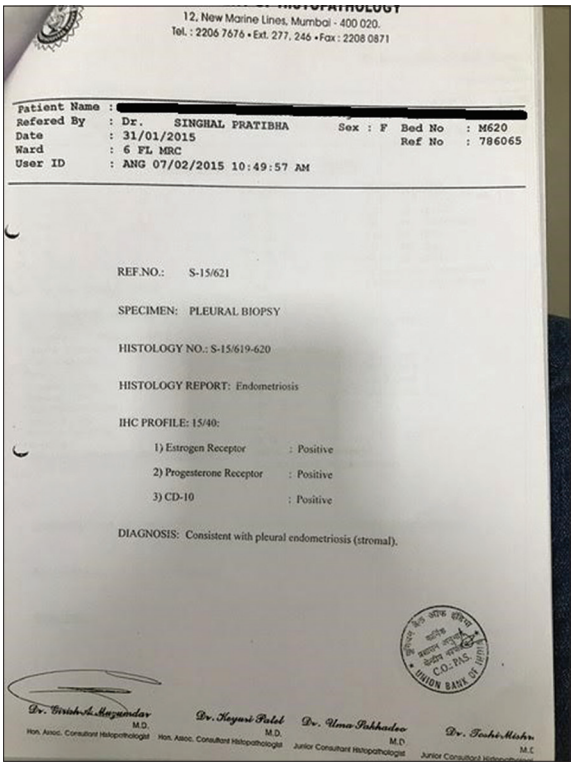


Figure 3: (a) International Classification of Diseases - lateral view. (b) International Classification of Diseases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Patient Name : [redacted]
Refered By : Dr. SINGHAL PRATIBHA Sex : F Bed No : M620
Date : 31/01/2015 Ref No : 786065
Ward : 6 FL MRC
User ID : ANG 07/02/2015 10:49:57 AM

REF.NO.: S-15/621

SPECIMEN: PLEURAL BIOPSY

HISTOLOGY NO.: S-15/619-620

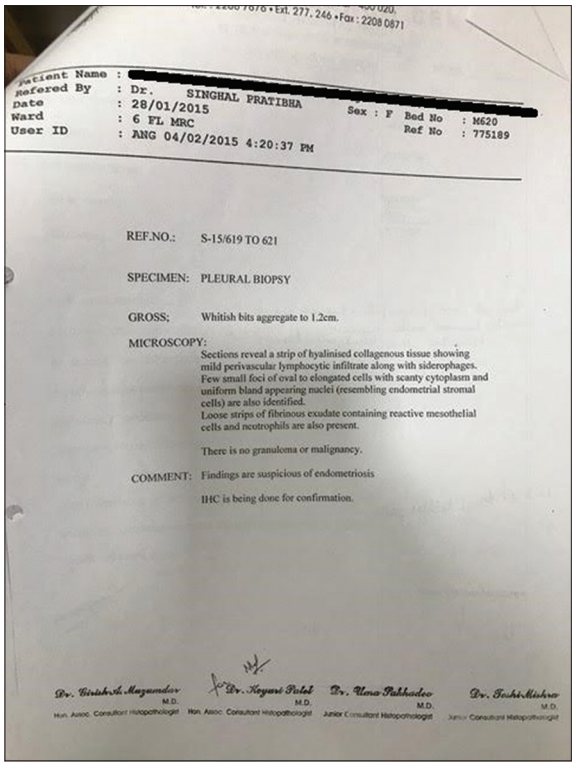
HISTOLOGY REPORT: Endometriosis

IHC PROFILE: 15/40:

1) Estrogen Receptor : Positive
2) Progesterone Receptor : Positive
3) CD-10 : Positive

DIAGNOSIS: Consistent with pleural endometriosis (stromal).

Figure 5: Immunohistochemical report


Patient Name : [redacted]
Refered By : Dr. SINGHAL PRATIBHA Sex : F Bed No : M620
Date : 28/01/2015 Ref No : 775189
Ward : 6 FL MRC
User ID : ANG 04/02/2015 4:20:37 PM

REF.NO.: S-15/619 TO 621

SPECIMEN: PLEURAL BIOPSY

GROSS: Whitish bits aggregate to 1.2cm.

MICROSCOPY:
Sections reveal a strip of hyalinised collagenous tissue showing mild perivascular lymphocytic infiltrate along with siderophages. Few small foci of oval to elongated cells with scanty cytoplasm and uniform bland appearing nuclei (resembling endometrial stromal cells) are also identified.
Loose strips of fibrinous exudate containing reactive mesothelial cells and neutrophils are also present.

There is no granuloma or malignancy.

COMMENT: Findings are suspicious of endometriosis

IHC is being done for confirmation.

Figure 4: Histopathology report

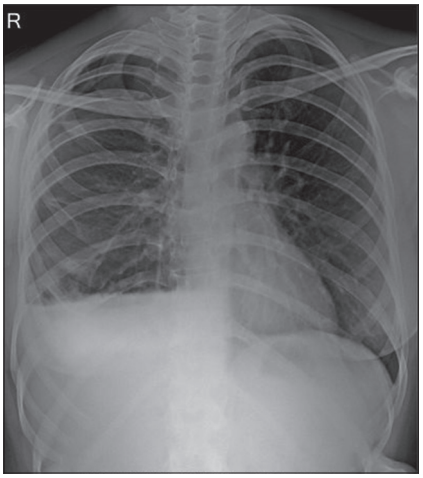


Figure 6: Persisting right pleural effusion

endometriosis, such as dysmenorrhea and dyspareunia. The detection of catamenial pneumothorax with the intercostal drainage during her menstrual cycle led to the diagnosis of pulmonary endometriosis, which in this patient was mostly only pleural and on the right side. Literature review does suggest that pleural endometriosis is more frequent on the right side,[6] as seen in this patient. Most of the cases of pleural endometriosis are associated with severe pelvic endometriosis[7] though this patient never complained about it, and the presence was detected only at the time C-section. The literature is still inconclusive about the best treatment strategy in the presence of TES in infertile young patients. Pleurodesis, especially in younger patients, should always be carried out concomitantly with VATS exploration. This way, an accurate diagnosis and treatment can be instituted and recurrences avoided. In older women and those not desirous of pregnancy, hysterectomy and bilateral salpingo-oophorectomy (BSO) is the treatment of last resort when other options have failed. Hence, a multidisciplinary approach by thoracic surgeon and gynecologist carries the highest chance of making an accurate diagnosis and providing the appropriate treatment strategies,[8] as seen in this case. Retrospectively, the decision to plan IVF-ET was the right choice for this

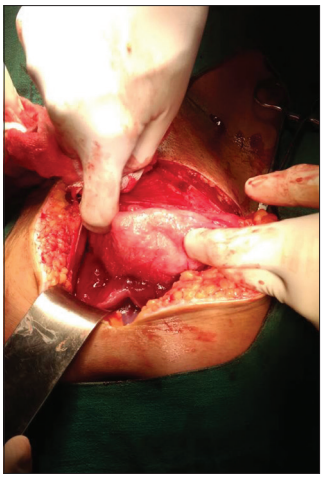


Figure 7: Intraoperative findings

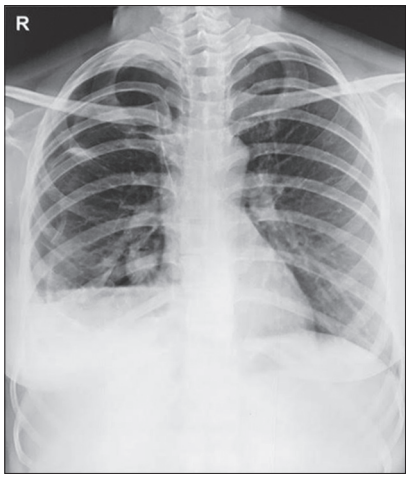


Figure 8: X-ray chest postoperative

patient and interestingly during the stimulation for IVF and subsequent transfer, and 9 months of pregnancy, the patient did not have any deterioration of the disease.

Literature review

To date, data are limited on the association between pelvic endometriosis and fertility status in patients with TES. In a recent series (2017) of 16 patients of TES by Ottolina *et al*.,[9] the study supported the presence of a strong association between catamenial pneumothorax and pelvic endometriosis. Pelvic endometriosis was diagnosed in 9 (56.3%) of the patients with TES. However, six patients did not undergo laparoscopic procedure to confirm or exclude the disease. Seven of the affected patients (77.8%) had moderate-to-severe disease (Stage III–IV), four conceived spontaneously after laparoscopic surgery, and all patients who did not have pelvic endometriosis conceived spontaneously. Although the study concludes that women affected by TES should be reassured about their childbearing capacity, a large study should be undertaken to definitely clarify this aspect.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initial will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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