

Therapeutic Challenge in Coexisting Infertility Pathologies; A Rare Case Report of Concurrence of Endometriosis, Adenomyosis and Tuberculosis

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ABSTRACT

One may find a real challenge in diagnosis and management of women with coexisting infertility pathologies. A rare case report of concurrence of endometriosis, adenomyosis and tuberculosis is discussed here. Appropriate clinical judgment and infertility treatment resulted in optimal outcome.

Keywords

Coexisting, Pathologies, Tuberculosis, Adenomyosis, Endometriosis.

Introduction

Infertility is one of the commonest presenting features of endometriosis as well as pelvic tuberculosis. One has to keep high index of suspicion as concurrence of these pathologies along with adenomyosis has been disregarded. Unusual clinical pictures of these entities confounded by diagnostic modalities usually cause management dilemma and may hamper surgical as well as infertility treatment outcomes [1].

Endometriosis is prevalent in 25-40% of infertile women. Adenomyosis is frequently diagnosed in women with endometriosis, but its role in subfertility is not precisely described due to lack of sufficient data. There exists some indirect evidence from cases studies of infertile women, who conceived after being treated for adenomyosis [2]. Possible mechanisms proposed for causing subfertility in women with endometriosis and adenomyosis are altered gene expression, disruption of endo-myometrial interphase, altered endometrial receptivity, and decreased ovarian reserve and associated concomitant factors like leiomyoma, endometrial polyp, and hyperplasia.

Tuberculosis, a hyperendemic disease of India, account for one fifth

of total global burden [1]. Majority of the cases are diagnosed after proper clinical evaluation along with appropriate investigations i.e. microbiological, imaging and histopathological. However, in few cases one may feel a real challenge. Empirical treatment of tuberculosis is justified in such cases with high index of suspicion, but should not be equated to indiscriminate use of antitubercular drugs [3].

An unusual case of concomitant infertility pathologies i.e. endometriosis, adenomyosis and tuberculosis is reported here. This kind of report is not evident in literature so far. Diagnosis was a real challenge in index case as despite intensive investigations and repeated relook surgeries, nothing was contributory. At last management on the basis of clinical judgment followed by appropriate fertility treatment resulted in successful pregnancy.

Case Report

A 33-year-old female with primary infertility of eight years presented with complaints of severe lower pain abdomen and fever for past two months. Her complaints started after she had undergone lapro-hysteroscopy for bilateral endometriotic cysts. There were bilateral endometriotic cysts of 5-6 cm along with grossly dilated fallopian tubes on both sides, which were adherent to endometriotic masses. Uterus was buried under the hydrosalpinges. Endometriotic cysts were removed along with salpingostomy of bilateral fallopian tubes. Hysteroscopy revealed

normal findings and no evidence of tuberculosis was observed. She was given injection leuprolide depot 3.75 mg intramuscular in her postoperative period. Histopathological examination of tissues confirmed endometriosis. The polymerase chain reaction, AFB staining and culture of endometria tissue showed no evidence of tuberculosis. She started developing high grade fever after two days of surgery and her fever persisted despite broad spectrum antibiotics coverage and there was nothing conclusive on sonography. The blood investigations did not reveal any sepsis. On relook laparoscopy after 6 weeks of first surgery, nothing could be visualized clearly because of dense adhesions between omentum, peritoneum, uterus and bowel.

At her first visit at the clinic, she was febrile. On per vaginal examination uterus was 16 weeks size. Ultrasonography revealed enlarged uterus with ill-defined endometrium and bilateral adnexal masses of 10x12 cm each with encysted fluid in pouch of Douglas (POD) [Image-1]. The blood tests for sepsis were negative, CA-125 was 38 miu/l and AMH was 0.67 ng/l. Patient was counselled for antitubercular treatment (ATT) on clinical grounds but she denied in view of lack of evidence. She was lost to follow up and thereafter revisited after one year later with persistence of fever, dysmenorrhea and lower abdominal pain. Patient was sick looking, uterus was grossly enlarged with big bilateral adnexal masses and encysted fluid in the POD. Repeat investigations again did not show any evidence of sepsis or tuberculosis.



Figure 1: Persisting bilateral adnexal masses.

She was started on ATT after proper counselling. Fever and dysmenorrhea subsided after intensive phase of the treatment but masses and fluid persisted. After complete course of ATT there was significant reduction in the adnexal masses. She was given option of in-vitro-fertilization (IVF) with ovum donation for infertility management. Before IVF, she was given three doses of leuprolide at an interval of 28 days for endometrial preparation. A great difficulty was encountered to negotiate the internal os and a flexible outer metallic catheter was used. Endometrium was built up with estrogen replacement and two good quality embryos were transferred. β hCG on day 14 was 237 miu/l and pregnancy continued till 37 weeks. She had placenta previa, mild IUGR and breech presentation. She was taken for caesarean and dense adhesions between uterus, omentum and bowel were observed. Lower segment was exceptionally thick and baby was delivered as breech with great difficulty. She had postpartum haemorrhage

which was managed with oxytocics. The postoperative period was uneventful and she was discharged on day 7 with healthy baby.

Discussion

Coexisting factors like tuberculosis, endometriosis and adenomyosis have greater impact on infertility and in some cases may be difficult to rectify. These patients may present with big adnexal masses mimicking ovarian malignancy and diagnosis has been confirmed after subjecting the specimen of oophorectomy/salpingectomy for histopathology [4]. Possible hypothesis for this may be exaggerated response of tuberculosis due to the release of proinflammatory mediators in endometriosis. Index case also had huge adnexal masses non responsive to any treatment but unlike other cases where investigations were contributory for the diagnosis of tuberculosis, none was conclusive in this patient. Empirical use of ATT was justified as the patient became severely ill and there was high index of suspicion because of clinical manifestations consistent with tuberculosis. With drastic clinical improvement after intensive phase decision to continue the treatment for 9 months was taken with close follow up.

The association of tuberculosis with adenomyosis is reported rare. Persisting foci of tuberculosis in the adenomyotic tissue may result in miliary spread [5]. Adenomyosis has detrimental effect on conception and there is increased risk of abortion [6]. Associated endometriosis further may add to increased morbidity and worsen the prognosis another challenge in this patient was grossly enlarged adenomyotic uterus. In accord with literature pre IVF leuprolide acetate given to index case resulted in successful pregnancy. Zhou et al. observed that 3-6 cycles of GnRh agonist before IVF causes significant decrease in uterine volume with better implantation rate [7]. Significant improvement in conception rate in frozen embryo cycle also after long pituitary downregulation with agonist has been reported in adenomyosis [8]. Direct antiproliferative effect within myometrium, reduced inflammatory reaction, reduced angiogenesis and hypoestrogenic state are thought to be responsible for improved outcome.

Conclusion

One thing to emphasize is that clinical judgement is supreme and there is no role of multiple relook surgeries. Management dilemma and subfertility are no doubt major challenges in such cases, one should be cautious to handle problems during caesarean section also. Suppression of adenomyosis, endometrial evaluation and cavity negotiation are other important concerns to be addressed.

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