Concomitant Lung Abscess and Endometritis in a Woman with Turner Syndrome in Postpartum Period in COVID-19 Era: A Case Report

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ABSTRACT

Objective: To report a rare case of Turner syndrome (TS) in postpartum period in COVID-19 era.

Case Report: A 27-year-old woman with TS and twin pregnancy referred for preterm premature rupture of membranes (PPROM) and severe anemia. Hysterotomy was done in this case and after that, she presented a persistent fever. PCR result for COVID-19 was negative. She had hemoptysis and anti-tuberculosis drugs were given. Lung abscess was diagnosed and right lobectomy was performed. Regardless of it, the fever continued and hysterectomy were carried out. Despite of this, fever continued. Finally, after 57 days, the patient was discharged. The patient was followed up for three months and she was afebrile and had no cough.

Conclusion: Women with TS are cases with multiple problems and it is necessary to mitigate them. In COVID-19 era, some of postpartum complications in these patients are like to COVID-19 symptoms and make their management difficult. Clinicians should consider life-threatening risks in these patients before recommending and using assisted reproductive techniques (ART), especially in more than one embryo transfer.

Keywords: Aspiration Pneumonia; COVID-19; Fertility Preservation; Lung Abscess; Turner syndrome

Introduction

Turner syndrome (TS) is the most common female sex chromosome disorder with an incidence of 1 in 2,000 to 1 in 2,500 live female birth [1]. TS is characterized by a complete or partial absence of one X chromosome. The most frequent chromosome constitution is 45X0, approximately half of patients have a mosaic chromosome complement, the most common being 45X0/46XX (15%) and 46Xq or 46Xp deletions (6%). The typical clinical features of TS are short stature, square appearance, webbed neck, characteristic unusual facies, low posterior hairline, broad chest with widely spaced nipples, and a “shield” chest [2]. Abnormalities in these patients including, cardiovascular disease which is highly prevalent and a major cause of early death [3], liver dysfunction, metabolic disorders, hypothyroidism, hearing loss, ocular abnormalities, skeletal anomalies, primary ovarian insufficiency and dysgenesis. The prevalence of these risks varies with phenotype and karyotype [4].

Most women with TS (95%-98%) are infertile due to gonadal dysgenesis, so natural pregnancies and live births
are rare (2). Despite this, detectable serum antimullerian hormone (AMH) levels, normal serum follicle-stimulating hormone (FSH) levels, and mosaic karyotype are predictors of the presence of primordial follicles [5]. After a literature search, clinical articles reporting postpartum TS complications in COVID-19 era were not found. Therefore, it is necessary to recognize and rule out TS complications from COVID-19 [6]. In this paper, we report adverse outcomes of a TS case with twin pregnancy in COVID-19 era.

Case Presentation

A 27-year-old mosaic (45X0/46XX) TS with twin pregnancy referred to AL Zahra Hospital with preterm premature rupture of membranes (PPROM) and severe anemia without any symptom of COVID-19. Her twin pregnancy had achieved via in vitro fertilization (IVF) using donated oocytes. She had a history of diabetes mellitus type1, epilepsy, and hypothyroidism. Sonography showed two embryos with gestational age of 19 weeks; one embryo had echogenic bowels and bilateral choroid plexus cyst. There was a huge retroplacental hemorrhage. Initial laboratory assessment showed severe anemia (Hb:6.6), leukocytosis (15300 with lymphocytes: 5%), increased C-reactive protein (63mg/dl), and increased erythrocyte sedimentation rate (ESR) (85mg/dl). She was given misoprostol (PGE1) for ripening the cervix and termination the pregnancy and also received packed cell but suddenly, she presented tachycardia (PR:140), tachypnea (RR:28), dyspnea, and decreased O2 saturation (85%). She became ready for operating room for hysterotomy. In her procedure, there was about 1 litre blood and clot in the uterus and two embryos were delivered. Uterine atonia was happened and controlled by misoprostol E1(1000µ), bilateral uterine artery ligation, and using some square sutures on the uterus. At the end of operation, there was decreased O2 saturation up to 47% and the patient could not be extubated. Portable chest x ray (CXR) showed bilateral infiltrative lung involvement so, she was transferred to intensive care unit (ICU). Chest computed tomography (CT) scan revealed no pulmonary thromboemboli (PTE). There was right pleural effusion, aspiration pneumonia, and indeterminate findings for COVID-19 (Figure 1), but PCR result for COVID-19 was negative, regardless of it, hydroxychloroquine prescribed for 10 days. After surgery, she presented a temperature up to 39° continued for 15 days. Laboratory assessments showed increased CRP (87mg/dl). On fever work up, the lung was the source of fever and other physical examinations were normal. She was extubated after 16 days of ventilation and given empiric broad antibiotics, including meropenem, vancomycin, linezolid, and caspofungine. After 3-day free of fever, again she showed a fever 20 days after admission, a pulse rate of 120/min, and blood pressure of 130/75mmHg. Abdominal examination was normal and the uterus was 3 cm above pubic symphysis with firm contour. Two days after new onset of fever, hemoptysis occurred. Chest CT showed thick wall cavitory lesion in posterior aspect of right upper lobe and superior segment of right lower lobe suggested active posterior primary tuberculosis (TB) (Figure 2). Consultant with thorax surgeon and infectious diseases specialist was performed and recommended to thoracotomy due to large cavity in lung. She was prescribed etambutol and streptomycine as anti-TB drugs. Lobectomy and irrigation were performed and a chest tube was inserted. In pathology report, right lung lobectomy was compatible with abscess formation.

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Two days after thoracotomy, she showed cardiopulmonary arrest and cardiopulmonary resuscitation (CPR) was performed. Chest CT revealed right side empyema. There was no PTE (Figure 3). Regardless of thoracotomy, temperature up to 39° continued. The blood culture report indicated gram positive cocci and sputum culture revealed pseudomonas and klebsiella resistance to meropenem. Echocardiography result revealed no endocarditis or any other pathology. Eight days after thoracotomy, the patient was extubated. The report of BK was negative and anti-TB drugs were discontinued. Due to continuing fever, consultant with thorax, lung, infectious, and cardiac services was performed and they recommended an exploratory laparotomy. On 22 days after thoracotomy, she underwent diagnostic curettage. Endometrial biopsy report revealed necrotic endometrium, so laparotomy with midline incision was carried out. The uterus was soft and foggy on palpation, then after the bladder flap was dissected free from uterus and cervix, with mild traction on uterus, the previous hysterotomy incision was abruptly interrupted. For this reason, and due to frozen section report, hysterectomy and bilateral salpingectomy were performed. There were two streaky ovaries in each pelvic side in which preserved. She was put on tazocine and caspofungine.

Pathology report revealed necrotic, degenerative endometrium, plasma cell infiltration that were consistent with chronic endometritis (Figure 4). Dispite hysterectomy, a low-grade fever (up to 38.5°) continued. Chest CT was done and showed loculated pleural effusion around right lower lobe containing discrete air bubbles in lobectomy site and indeterminate finding for COVID-19. Interventional radiology consultant was done but it was impossible to tap this loculated fluid and counselling to thorax service was recommended. Again, PCR for COVID-19 was negative. Thorax consultant suggested that she could be discharged with cefexime and clindamycin without any surgery and should be visited in short intervals (Figure 5).

Finally, after 57 days, the patient was discharged with temperature of 38° and pulse rate of 120/min. On 3 follow up visits, (on 3 days, one and two weeks after discharge), there was no abnormality in examination except tachycardia (PR:120/min). Her serum Hb level was 10 mg/dl. The patient complained of infrequent cough. She was symptom free 3 months after follow up.
**Discussion**

We described a woman with mosaic TS who got twin pregnancy following IVF with oocyte donation. In addition to cardiopulmonary arrest, she had some postpartum complications such as prolonged fever, aspiration pneumonia, lung abscess, and endometritis, which were in line syndrome of COVID-19. According to the literature, many studies have been carried out on IVF in TS, few studies have focused on postpartum events [7,8]. Aspiration of small amounts of oropharyngeal secretions is common even to healthy individuals and usually resolves without detectable sequelae. Sensitive tests show that at least one-half of healthy adults’ aspirate during sleep. Thus, most episodes of aspiration are subclinical and/or rapidly resolve without clinical manifestations. The small fraction of aspiration events that do proceed to clinically manifest disease appear to depend upon the volume and contents of the inoculum as well as failures of host defense mechanisms [9]. There are many factors that was made aspiration pneumonia in our case, such as, emergency hysterotomy and anesthesia. But one of the causes of developing lung abscess and prolonged fever following aspiration pneumonia may be attributable to defects of immune responses in these patients.

Moreover, TS may be associated with immune abnormalities. TS patients have been reported to have immune alterations in T cell and immunoglobulin subsets. These include decreased levels of circulation T and B lymphocytes, reduced levels of serum IgG and IgM, and increased IgA [10]. However, other studies did not find major immunological deficiencies in TS subjects [11]. A total of 1169 unique genes showed different expression in TS peripheral blood mononuclear cells (PBMCs) including 35 on the X chromosome. In particular, ubiquitously transcribed tetratricopeptide repeated on chromosome X (UTX) was among the top 10 X-linked genes with the largest decrease in expression and the only gene among these candidates that escapes X inactivation. TS patient immune cells express decreased UTX, a Histone-Modifying Enzyme. UTX Deficiency in T cells prevents T follicular helper (Tfh) differentiation and eradication of chronic infection. TS patients have decreased Tfh cells [10]. In our case, there were aspiration pneumonia, pneumonitis, endometritis, and prolonged fever. It is supposed that lower immunoglobulins, immune cell deficiency and failure of defense mechanisms in women with TS predispose them to these situations. Therefore, treatment with intravenous immunoglobulins (IVIG) could be effective. According to study of Bradly J et al., decreased Tfh cells in TS suggested impaired response to viruses and vaccines [10]. We’re living in COVID-19 era and TS patients are a part of the society, thus, it is recommended to design a study to evaluate which type of vaccine and how many doses of vaccines are needed to vaccinate these women.

Due to inherent cardiovascular abnormalities associated with TS, these women are at the increased risk of dying from aortic dissection or rupture owing to the increased cardiovascular demands of pregnancy. These demands are further increased in multiple gestations, which are often an unfortunate consequence of ART. For this reason, it is imperative that any woman with TS who is undergoing treatment with IVF or donor oocyte has only a single embryo transferred [12] which is in line with Bouet et al’s study [13]. Due to small uterine size of women with TS even after hormonal therapy, Folsom et al also recommended that only one embryo should be transferred per IVF cycle to prevent undue complications [14]. Data of Calanchini et al. confirmed that IVF–oocyte donation with double embryo transfer is associated with miscarriages and poor maternal and fetal outcomes [15], so in our case if one embryo had transferred, maybe PPROM and the following complications wouldn’t have happened.

In sum, any pregnancy in woman with TS must be considered as high risk of innately cardiovascular risk related to structural cardiac anomalies that this population predisposed to. However, TS is not an absolute contraindication to pregnancy, but there are no screening methods to divide the potential population into those individuals whose underlying cardiovascular risks are too high to allow pregnancy and those who might reasonably be monitored and guided by a skilled perinatologist through a high-risk pregnancy [16]. Despite these risks for a pregnancy, there is a tendency to oocyte donation to achieve a pregnancy, so, it seems logical to transfer one embryo instead of two, to mitigate pregnancy and postpartum complications. Finally, as the best option, use of gestational surrogate is recommended for women considering use of donor eggs for eliminating these risks and to be a biological parenting.

**Disclosure**

The authors declare no conflict of interest.

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