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Case Report

A Rare Case of Shared Delusion of Pregnancy in a Couple Undergoing Fertility Treatment

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ABSTRACT

Pseudocyesis or phantom pregnancy is a false belief of being pregnant that is associated with objective signs and reported symptoms of pregnancy, which may include abdominal enlargement, reduced menstrual flow, amenorrhea, subjective sensation of fetal movement, nausea, breast engorgement and secretions, and labor. Though it is a very rare condition, it is often seen in women undergoing fertility treatment in developing countries. Isolated pseudocyesis in women and sometimes men have been reported before, but there are very few reports of shared delusions of pregnancy by a couple. No such report has ever been made before from Nepal. Therefore, this case report seeks to associate psychiatric/psychological, gynecological, and neuroendocrine mechanisms leading to the emergence of shared delusion of pregnancy (folie à deux) in a Nepali couple undergoing artificial reproductive techniques for the management of infertility. In this case, the couple firmly believed in their pregnancy against all medical evidence.

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INTRODUCTION

Infertility, a huge taboo in traditional Asian societies, predisposes one to psychological trauma, anxiety, stress, depression, and psychosis. Phantom pregnancy or pseudocyesis is a rare psychological condition sometimes seen in women suffering from infertility that has captured the attention of both clinicians and non-clinicians for centuries. It has been featured in multiple books, movies, television shows, and psychiatric studies. In this rare clinical syndrome, a nonpregnant woman believes she is pregnant and exhibits signs and symptoms of pregnancy in the background of a negative urine or blood pregnancy test. Medical literature describes pseudocyesis as a very rare condition, with a prevalence of 1-6 cases per 22,000 live births.[1]

Pseudocyesis is more prevalent in traditionally patriarchal cultures, where women are often divorced if unable to bear offspring. Infertility in such societies is often attributed to the woman, which results in her experiencing substantial distress and discrimination predisposingher to delusions of pregnancy.[2] There are a few case reports of pseudocyesis from Nepal. However, here we present a case of pseudocyesis leading to folie a deux, a shared delusion of pregnancy between husband and wife. Folie a deux is a term coined by Lasegue and Falret in 1877 meaning shared psychotic disorder.[3] In this condition, a member (wife in this case) develops the delusion first and is likely the dominant member in a close relationship with a more suggestible person (the husband)

who alsodevelops the delusion.[4] Herein, we describe such a case in which a married couple undergoing in vitro fertilization for infertility treatment shared delusions of pregnancy in spite of substantial medical evidence to the contrary.

 # CASE REPORT

Thirty-nine-year-old regularly menstruating female presented to Vatsalya Natural Health Care on May 2, 2019, with complaints of infertility for 5 years. According to the patient, she had two previous conceptions at the age of 23 and 26 years but had undergone induced termination of pregnancy with 200 mg mifepristone followed by 4 tablets of misoprostol (200 micrograms each) at 8 and 8+4 weeks of gestation respectively. The documents showed an ultrasound indicating live intrauterine pregnancy prior to termination. Both abortions were executed under the supervision of a nurse at Meristop Safe Abortion Center, Kathmandu, and led to no complications. The patient stated that the reason for terminating the pregnancy was that she was studying at that time. At the time of presentation at Vatsalya, the patient mentioned that she had undergone two previous cycles of in vitro fertilization, once at Creators IVF and once at Alka IVF. The first cycle at Creators led to the formation of two 5AA blastocysts which were implanted together in a fresh embryo transfer cycle but did not result in pregnancy. The second cycle at Alka IVF led to the formation of 5 8I day 3 embryos. The frozen cycle was done and 3 8I embryos were transferred on the first attempt but the β-hcg test came out negative. On the next attempt, 2 8I were transferred,but pregnancy did not result.

At Vatsalya, a hysteroscopy was done prior to initiating the IVF cycle and a bulky uterus with a normal uterine cavity was noted. Ovarian stimulation was done with 150 iu Gonal F (FSH) for 20 days. On day 6 the follicular size was noted to be 16 mm, 14 mm,13 mm, 13.5 mm, 16 mm on the left and14 mm, 11mm, and 14.5 mm on the right side, and 75 IU HMG was added from day 6-day 10. Also, Inj cetrorelix 0.25 mg was given from days 6-10. Triggering was done with 5000 units of HCG and 5M2 oocytes were obtained which were fertilized leading to the formation of 2 blastocysts: 4AA, 4AA. The two embryos were implanted 5 days after ovum pickup as fresh embryo transfer, and β-hcg was done after 14 days which was negative.

The result was communicated to the patient, but the patient came to the clinic after a week stating that her home pregnancy test was positive. A repeat β-hcg was done at the clinic whichagain came back negative. It was explained that the home pregnancy test may have been faultyand the patient and her husband were counseled regarding the results. The patient stated that the results are wrong and that she hasn't gotten her period since the transfer, and as her periods are always on time, she must be pregnant. It was explained that hormonal therapy can cause a delayin periods and it will still come. Both the patient and husband did not accept the explanation.

The patient went home and was contacted via telephone one week later to follow up. Both the husband and wife claimed that the pregnancy was going well on the phone stating the wife now has morning sickness. The husband hung up the call when it was explained that the chances of being pregnant are minimal. The doctor requested a follow-up visit, but the patient did not come and the clinic number was blocked.

One month after the phone call, the patient's husband came to the clinic in an upset state to discuss his wife's condition. He stated that his wife had aborted again and had heavy bleeding.He wanted to know how long before they could start another cycle. The doctor requested a conference with other family members, but the husband said that they had been doing infertilitytreatment secretly and did not want to involve other family members.

The doctor requested a follow-up with the wife and decided to involve a mental health counselor, however, the clinic was closed due to covid 19 lockdown for 3 months and the patient was lost to follow-up (March 2020-May 2020). In December 2020 when the clinic was fully operational again, the patient was contacted but the husband stated that they were planning to go to IndiraIVF in India in the upcoming week to pursue further fertility treatment.

DISCUSSION

This patient's history showed the onset of pseudocyesis in her late 30s, which is considered very late for a woman to be childless in eastern cultures. By this time, women often suffer from threats of divorce, domestic violence, and abandonment. The consequences of infertility for couples include social isolation, economic deprivation stigma, and discrimination.[5] A study done by WOREC in rural areas of Nepal showed that Infertile couples have adverse psychological effects and express profound effects on mental health. Couples with no children shared that they were suffering from fear, anxiety, stress, and depression resulting from infertility, with women more affected than men.[6] Infertility can thus have serious implications on both the psychological well-being and social status of couples living in developingcountries.[7]

Thus, it seems only logical that the pressures to bear a child may predispose to a delusion of pregnancy in developing patriarchal societies like Nepal. The motives underlying pseudocyesis include the need to secure a position in society as a child bearer, thereby helping the husband's family to continue their lineage. Such delusions are usually preceded by psychosocial stress orsuch as divorce, abortion, miscarriage, or failed IVF. Pseudocyesis or "phantom" or "false" pregnancy involves psycho neuro-endocrine control of the gonads such that symptoms of pregnancy such as amenorrhea, abdominal distention, breast engorgement, enlargement of the uterus, perception of fetal movements may be present even in the absence of actual pregnancy. Itoften presents with varying severity and could start from transitory fears of being infertile leading topathological changes in neuroendocrine functions.[8]

Data support the notion that pseudocyesis women may have increased sympathetic nervous system activity, dysfunction of central nervous system catecholaminergic pathways, and decreased steroid feedback inhibition of gonadotropin-releasing hormone.[9] It also has been established that the activation of the amygdala, mediated through the amygdalofugal pathways and the stria terminal, can affect hypothalamic-releasing factors that control the release of ACTH and the pituitary gonadotropic hormones. The significant inputs from the orbital frontal cortex and anterior cingulate region into specific amygdaloid nuclei indicate that cognitive-affective processes and real or anticipated changes in personal bonding could influence the amygdala's modulation of hypothalamic function.[10] Following this assumption, Persinger MA found thatwomen who develop pseudocyesis also displayed

significant elevations in temporal lobe signs (showing more incidence of complex partial epilepsy, interictal-like signs, memory blanks, automatic behaviors, depersonalization, olfactory sensation, and increased abdominal awareness) with particular involvement of putative right-hemispheric processes.[11]

In this case, the delayed periods in this patient and the morning sickness may have been due to such neuroendocrine mechanisms. However, they may also be attributed to hormonal therapies used during IVF which can cause irregularity of cycles in some patients. Whatever the reason, the delay in periods, positively reinforced the couple's delusion that the wife was pregnant, so much so that they completely disregarded the medical evidence presented to them. The couple even confused menstrual periods with abortion and expressed grief at the pregnancy loss.

The duo presented in the index case study fit the diagnosis of folie-à-deux as well, meaning a shared delusion. Folie a deux is a condition described by Lasegue and Falret in which the dominant member (wife in this case) develops the delusion first and "induces" delusions in the more suggestible person (the husband). As the husband mentioned that his family does not know about them undergoing fertility treatment, their isolation from others may have reinforced their development of shared psychosis. Usually, the person who develops delusions second (in this case the husband) is frequently less intelligent, more gullible, more passive, or more lacking in self-esteem than the person in the primary case. If the pair separates, the second person may abandon the delusion, but this outcome is not seen uniformly.[12] In our scenario, we were notable to follow up on the psychiatric evaluation of the patients and hence not able to observe the outcome of separation.

The management of pseudocyesis is multidisciplinary, including psychiatrists, gynecologists, and psychologists. The goal of treatment is to help patients perceive the meaning of their symptoms and to help resolve the associated stressors. Psychotherapies can be combined with medications like an antidepressant and antipsychotic medications depending on the associated comorbidities.[13] Unfortunately, in our case we were not able to successfully administer mental health counseling and treatment to the couple, as they were lost to follow-up due to covid-19. Moreover, the couple later opted to change medical providers.

The authors believe that mental health counseling should be an integral part of fertility treatment. If mental health issues are often discussed and patients are given support, it may prevent the reinforcement of delusions which stem from emotional stress.

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