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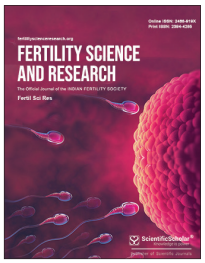


*Case Report*

# Infertile Couple with Single Ovary and Single Testis: Miracle Baby

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## ABSTRACT

Any gender with a single gonad can suffer from infertility due to various reasons. A married couple with single gonads: a 34-year-old male with single testis following orchidectomy and a 31-year-old female with single ovary following unilateral ovariotomy presented for infertility management. The female partner had a low ovarian reserve, and the male partner's semen analysis was low rapidly progressive motility. They could not achieve pregnancy naturally for two years. The female partner was treated with tablet Dehydro-Epiandrosterone Sulphate (DHEA), with three cycles of ovulation induction and timed intercourse and later three cycles of intrauterine insemination (IUI). The first cycle of IUI resulted in a biochemical pregnancy. She had spontaneous conception during the cycle of downregulation for in vitro fertilisation (IVF) with an injection of leuprolide on day 21. She delivered a live child of 2.7 kg without any anomalies, and the milestones were normal.

**Keywords**: Couple with single testis and single ovary, unilateral ovariotomy, mucinous cystadenoma, unilateral orchidectomy, trauma, low ovarian reserve, IUI

## INTRODUCTION

Infertility is not only a personal problem but is also a social, psychological and medical problem. It has become a public health issue as it is reported to affect one in four couples. Primary infertility is more common than secondary infertility, and the female factor has been cited to be the main contributory cause.[1] The exact contribution of male and female factors for infertility varies with age, habits, geographical distribution and the type of facility where the study are conducted.[2] Some studies reported 50% of etiological factors attributed to males [3], but few others reported only 20%.[1] There are various reasons why a male undergoes orchidectomy at a young age, like torsion testis, undescended testis, trauma and rarely malignancy. Most commonly, we encounter women with single functional ovaries following oophorectomy for torsion ovarian cysts or ovarian malignancy. It is rare to find the combination of a couple with a single ovary and testis and consulting for the problem of infertility. This case is shared for the above reason and also for presenting the cause of infertility in this couple and conception during management.

## CASE REPORT

A 31-year-old staff nurse who was married for one year was approached for fertility issues in August 2019. She has had regular cycles since menarche, once in 24–28 days and bleeding lasting

for 3–4 days. Her history was significant in that she underwent emergency laparotomy and left salpingo-ovariotomy for a torsion ovarian cyst in 2011 (ten years ago), and the histopathology was mucinous cystadenoma and normal fallopian tube. Her mother was diabetic and hypertensive, and there was no other significant family history.

On clinical examination, she was small built, and her body mass index (BMI) was 19 kg/m.[2] Her pulse was 82/min, blood pressure (BP) 108/75 mmHg, no palor and breasts and thyroid were normal. Abdominal examination revealed a midline sub-umbilical scar, which healed by secondary intention, and there was no hernia. Per speculum examination revealed a healthy nulliparous cervix and a healthy vagina. On bimanual examination, the uterus was retroverted and mobile and no tenderness or mass in the fornices. The transvaginal scan revealed a retroverted uterus with normal myometrium, and endometrial thickness was 8.4 mm. The left ovary was not visualised, and the right ovary was normal, with a small follicular cyst. She was advised to do semen analysis, haemogram, syphilis venereal disease research laboratory (S VDRL), Hepatitis B surface antigen (Hbs Ag), hepatitis C virus (HCV), day two hormonal profile, serum thyroid stimulating hormone (TSH) and hysterosalpingogram (HSG).

Her haemogram was normal, TSH was 2.2 mIU/ ml, *follicle-stimulating hormone* (FSH) was 8.5 mIU/ml, luteinising hormone (LH) 4.4 mIU/ml, prolactin 19ng/dl, S. testosterone was 23 ng/dl and S. *anti-müllerian hormone* (AMH) test was 0.9 ng/ml. HSG showed a patent fallopian tube. She was told about her low ovarian reserve and fertility problems and advised to take tablet DHEA 75 mg Dehydroepiandrostenidione, Richova (Manufacturer-LUPIN, India) once a day and do semen analysis, and her husband was to be brought for examination and counselling. The couple did not come for a year, and she did not take tablets of DHEA.

Semen analysis done in December 2020 showed a count of 40 million and motility of 75% with rapid progressive (A) at 10% and slow progressive (B) at 50%; at this time husband revealed a history of orchidectomy due to trauma. He was advised to take urology consultation and was advised to take tablet containing coenzyme Q 10, vitamin D3 and L-carnitine fumarate equivalent to L-carnitine 1 g, acetyl L-carnitine hydrochloride 0.5 g and fructose IP 1 g antioxidants in powder form Maxoza-L (Manufacturer SUN, INCA, India) once daily for three months. She was advised to take a tablet of DHEA and follow a fertile period. Her AMH was 1.2ng/dl in July 2021 (after three months of DHEA), and her antral follicle count (AFC) was 4. She underwent ovulation induction with 5 mg of letrozole for five days and injection r FSH 75 IU S.C In recombinant follicular stimulating Hormone (Follisurge, manufacturer-Inca's Pharmaceuticals, India) on days 7, 9 and 11; follicular monitoring was done, ovulation was documented and intrauterine insemination (IUI) was done in September 2021 as she did not conceive with timed intercourse which was followed for three cycles. Semen analysis at the time of IUI showed a count of 70 million, with Type A motility 28% and Type B motility 34% with few white blood cells (WBC). She had 38 days of amenorrhea and a positive pregnancy card test. Transvaginal ultrasound (TVS) showed a thickened endometrium of 18 mm with a doubtful gestational sac. Her beta HCG was 140 mIU/ml. She had bleeding per vaginum on the 42nd day, and beta HCG was 39 mIU/ml. A biochemical pregnancy failure was diagnosed, and she was reassured of pregnancy support during the next conception. She underwent two more cycles of IUI without any success, and later, hystero-laparoscopy was performed, which was normal with a patent right tube and normal right ovary without any adhesions. She has continued consuming tablet DHEA on her own for a further three months.

She was enrolled for the batch in vitro fertilisation (IVF) and was given an injection of leuprolide 3.75 mg Luprodex, (manufactured by BSV, India) i.m on 29.10.2022 on the 21st day of the cycle. She complained of nausea and vomiting and suspected pregnancy after ten days of taking injection, and her pregnancy card test was positive. Her β HCG was 39.82 mIU/ml on 31.10.22, 1317 m IU/ml on 6.11.22 and 5520 mIU/ml on 11.11.22. TVS performed on 18/11/22 showed a single intrauterine gestational sac with a crown-rump length (CRL) of 45 mm, corresponding to six weeks + one day. She was advised to continue taking tablets of folic acid and take a tablet of dydrogesterone 10 mg Duphastan, (manufactured by Aboott, India) twice daily for pregnancy support. She had regular antenatal care and had two hospitalisations for hyperemesis gravidorum. The foetal growth was normal, and she underwent elective caesarean section at 38 weeks due to cephalopelvic disproportion and a male baby with a birth weight of 2.7 kg was born. The baby had no congenital anomalies and had normal milestones.

## DISCUSSION

Ovarian cystectomy is most commonly undertaken for benign lesions and oophorectomy in emergencies like torsion and gangrene when it is not possible to undertake fertility-sparing surgery. The remaining ovary, after unilateral oophorectomy, is supposed to maintain the endocrine function. The review by Luisa MG *et al*. reported compensatory function in animal studies; in women also, there was a compensatory increase in ovarian volume; the biological effects are different and often have a negative impact on women's health, including cardiovascular, neurological and reproductive health.[3] The spontaneous pregnancy rate of 40–50% was reported following unilateral oophorectomy,

and there was no difference in pregnancy outcomes following Assisted Reproductive Technology (ART) in women with single ovaries compared to women having both ovaries.[4] However, decreased ovarian reserve was reported in a large retrospective study (1978–2015), which compared women with a single ovary to those with both ovaries.[5] A large multi-centre study reported a lower ovarian sensitivity index and lower oocyte yield in women with unilateral oophorectomy than controls after adjusting for confounding factors such as age.[6] The current patient had low ovarian reserve following unilateral oophorectomy and presented with infertility, though anovulation was not proved, and her cycles were shorter. Her AMH improved quantitatively with DHEA, and she ovulated with ovulation induction drugs.

Regarding male fertility, cryptorchidism is common in males who were born preterm, and more than 90% have oligospermia. The maturation of the hypothalamic-pituitary-gonadal axis is affected, which results in decreased sperm production even from the unilateral normally descended testis.[7] A cross-sectional case-control study of men with solitary testis, when compared to men with both testis, revealed high FSH and LH levels, though testosterone levels were compensated; they attributed this to pituitary strain and pituitary burn-out.[8] Even in unilateral testicular torsion, the contralateral testis dysfunction is reported.[9] Development of anti-sperm antibodies following testicular trauma is possible, and the effect of anti-sperm antibodies causing infertility is manifold, including reduced motility, inability to penetrate cervical mucus, abnormal acrosome reaction failure to fertilise, abnormal embryo development and implantation. Their capacity to reduce sperm motility causes sperm agglutination, impairs sperm's ability to penetrate the cervical mucus or inhibits sperm capacitation, the acrosome reaction and sperm-egg interaction, as well as the initial stages of embryo development.[10] Unilateral orchidectomy can decrease the fertility potential of a man. A comparative study involving 54 men with single testis of normal volume revealed oligospermia, irrespective of the cause of orchidectomy.[11]

In this couple, the cause may be a female factor with low ovarian reserve or anovulation as she conceived with ovulation induction initially, and later, when ovarian reserve normalised with DHEA, she had spontaneous conception. The administration of gonadotropin hormone-releasing hormone (GnRH) on day 21 did not affect the implanted embryo. The probability of implantation may be thought of as earlier than day 21 in this lady. Younis JS *et al*.[5] estimated the probability of conception to be 58% on day 12 and 5% on day 21, and the overall day-specific probability was thought to rise sharply seven days after the last menstrual period (LMP), reaching a maximum at 15 days and reaching zero by 25 days.[12] Spontaneous conception during downregulation is a known fact attributed to the flare-up phenomenon with a luteal phase rise of FSH and LH. The exact mechanism is not known, though it was hypothesised to occur due to improved crosstalk at the molecular level between the embryo and endometrium. Two spontaneous pregnancies in two downregulated IVF cycles with administration of leuprolide acetate 1 mg s.c daily from day 19 for 14 days were reported by Platteau P *et al*.[13] Luteal support is desirable for these pregnancies as they are at high risk of abortions because the luteolytic effect of GnRH lasts as long as nine weeks;[14] we have given luteal support for this woman based on this. GnRH directly or indirectly affects the endometrial stem cells and acts through its receptors (GnRH-R) present in the endometrium through the PI3K/Akt signalling cascade, which is involved in endometrial proliferation, differentiation and migration, and high doses or longer exposure can damage the endometrium.[15]

Fatima P *et al*. reported spontaneous pregnancies diagnosed on the 26th day after GnRH agonist, and a paediatrician followed up the children born out of the three pregnancies, and they were normal.[16] A large retrospective analysis of spontaneous pregnancies with GnRH downregulation over 17 years among 26,002 IVF cycles was analysed by Huan Wu *et al*.in China. They reported the incidence of pregnancies as 0.56%, and the long-term follow-up of the children did not show any adverse neurodevelopmental problems. [17]

## CONCLUSION

It is natural to expect infertility after losing the gonads in either couple or both, and an early evaluation and management are necessary. Spontaneous ovulation and conception can occur even in women with low ovarian reserve during the cycle of downregulation for ART, and this case illustrates that exclusion of pregnancy is necessary prior to commencing controlled ovarian stimulation when the woman does not get withdrawal bleeding. Couples can be reassured that there will not be any adverse effects on the children resulting from spontaneous conception after the administration of the GnRH in the luteal phase of the cycle.

### Ethical approval

Institutional Review Board approval is not required.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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### Conflicts of interest

There are no conflicts of interest.

### Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of AI-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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