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SUCCESSFUL TWIN PREGNANCY IN SWYER SYNDROME: CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Objective: To report a successful twin pregnancy in a patient with Swyer syndrome (46 XY pure gonadal dysgenesis). **Design:** Case report and review of literature. **Setting:** Infertility unit (Jindal Hospital, Meerut, Uttar Pradesh, (Delhi NCR) India. **Patient:** A female aged 31 years with complete 46 XY gonadal dysgenesis seeking fertility. **Interventions:** Intra Cytoplasmic Sperm Injection (ICSI) using donor eggs, fresh embryo transfer, caesarean delivery, followed by Laparoscopic gonadectomy. **Result (s):** Successful pregnancy and delivery of healthy twins following ICSI using donor oocytes & fresh embryo transfer in a patient of Swyer syndrome. **Conclusion:** Patients with Swyer syndrome can achieve a successful pregnancy with donor oocytes and ICSI, and deliver a healthy baby though it is a rare feat.

CASE REPORT

Rare case of twin pregnancy in a patient with Swyer syndrome (XY)

Background

Swyer syndrome, also called pure XY gonadal dysgenesis is a rare disorder of sex development (DSD) associated with infertility. It was first described by G. Swyer in 1955, when he found two cases of sex reversal which were different from usual cases of "male pseudohermaphroditism".[1] It has an incidence of around 1 in 80,000 births.[2]

It is characterized by a 46, XY karyotype, streak gonads which are completely undeveloped leading to azospermia and hypergonadotropic hypogonadism, female phenotype and normally developed Mullerian duct leading to normal uterus, fallopian tubes, and vagina. Sex of rearing is female because external genitalia is like a normal female. The diagnosis is often delayed and relies on the history of primary amenorrhea with normal female phenotype,

normal uterus and tubes, and 46 XY Karyotype.[3] In Swyer syndrome, early diagnosis is important because the gonads have a tendency of malignant transformation such as a dysgerminoma or gonadoblastoma in 15-35% cases.[4] This increases further with age, hence gonadectomy is recommended at the time of diagnosis.[5] This is then followed by hormone replacement therapy (HRT) to induce menarche and to maintain the long term bone health.[6]

Achieving a pregnancy in Swyer syndrome is a challenge because of gonadal failure and hypoplastic uterus. Conception is possible with assisted reproductive technology (ART) with ovum donation. Till date, only 29 patients of swyer syndrome with successful pregnancy have been reported worldwide.[16-43] Most of the deliveries were singleton only 10 patients had twin deliveries.

Out of these 29 patients there were three patients who delivered twice[22,23,43] and Michala et al reported three deliveries in the same patient of Swyer syndrome.[6,28]

We add to this record by reporting a successful twin pregnancy in Swyer syndrome.

Case report

A phenotypic female, aged 31 years, presented to infertility department of Jindal hospital, India with primary amenorrhoea and inability to conceive for 5 years. She had taken treatment from a general practitioner before reporting to us. She underwent a detailed clinical & investigational work up in view of primary amenorrhoea.

She was a tall, eunuchoid phenotypic female with a height of 178 cms, weighed 76 kg, body mass index (BMI) of 24 kg/m^2, tanner stage 3 breast development, grade 2 axillary hair, and normal female external genitalia with no evidence of clitoromegaly or virilism.

Pelvic ultrasound revealed uterus measuring 56mm X 25mm X 35mm with streak gonads. Hormonal profile revealed Hypergonadotrophic Hypogonadism with FSH 56 mIU/mL, LH 36 mIU/mL, E2 11pg/mL, prolactin 15ng/mL, testosterone 66ng/dL, TSH 3.6µIU/mL. Karyotype was XY, which clinched the diagnosis of Swyer syndrome.

Husband's semen analysis was normal.

The couple was counselled regarding the psychological implications of the condition and the need to induce menarche with Hormone Replacement Therapy (HRT) and gonadectomy to prevent future malignancy. The patient gave consent for HRT for initiating menarche but

refused for gonadectomy. She was started on cyclic HRT to induce menarche in the form of estradiol valerate 2mg 8 hourly for 10 days, increased to 4mg 8 hourly for 15 days to achieve a tri laminar endometrium followed by Medroxy-Progesterone Acetate 10mg (MPA) twice a day for 5 days for withdrawal bleeding. She achieved menarche after three months of HRT which was continued for next two months. Her uterine size was monitored sonographically increased to 65x32x36cm with HRT.

She gave consent for ART with donor egg programme for fertility as her chief complaint was inability to conceive. Endometrial preparation was done with HRT with Estradiol Valerate 4mg given 12 hourly (8 mg) for 8 days, further increased to 4 mg 8 hourly (12mg) for 9 days to achieve a tri-laminar endometrial thickness of 10.7mm. The anonymous egg donor was started on long agonist protocol with Human Menopausal Gonadotrophins (HMG) in a dose of 225 IU given IM from D 3 for 10 days. Seven mature eggs were retrieved, which were micro injected by Intra Cytoplasmic Sperm Injection (ICSI) using husband's sperm. Three Grade 1 embryos were obtained, out of which two Grade I fresh embryos were transferred on D 3. Pregnancy test (βHCG) done 14 days later did not reveal a pregnancy.

She underwent the second ICSI cycle with donor egg in the next cycle. Endometrium was prepared with HRT as done in the previous cycle. Eight mature eggs were retrieved from donor. ICSI was done with husband's sperm. Two Grade-I embryos were transferred in an easy embryo transfer setting with D 3 transfer. Pregnancy test done 14 days later was found to be positive (βHCG -526mIU/ml). Transvaginal ultrasound at six weeks confirmed the presence of two intrauterine gestational sacs with diamniotic dichorionic twin pregnancy.

The third trimester was fraught with difficulties with gestational hypertension (GH) and fetal growth retardation (FGR) setting in at 28 weeks of gestational which were managed conservatively by starting her on Tab Labetalol 100 mg twice a day. Antenatal steroids were given at 32 weeks to hasten lung maturity as she complained of preterm labour pains which was managed conservatively. Antenatal surveillance was done by regular ultrasound monitoring & CTG monitoring after 32 weeks. An elective caesarean was done at 37 weeks and healthy twins were born with a girl weighing 2400gm and a boy weighing 2225gm. The babies did not require any Neonatal Intensive Care Unit (NICU) admission. She even breast feed her infants till eight months. She later gave consent for gonadectomy and underwent a laparoscopic gonadectomy for the streak gonads 8 months after the delivery at our centre. She was put on HRT after gonadectomy to preserve her bone health.

DISCUSSION

Swyer syndrome is a rare disorder of sexual development (DSD) and occurs due to failure of development of sex glands (testes) in a genetic male (46 XY karyotype). It is pure gonadal dysgenesis. Pure gonadal dysgenesis is a condition in which gonads are not differentiated but external and internal genitalia develop normally. The National Organization for Rare Disorders (NORD) classifies Swyer syndrome as 46 gonadal dysgenesis expressed either in complete or incomplete form: 46XY complete gonadal dysgenesis, 46 XY pure gonadal dysgenensis, gonadal dysgenesis, XY female type.[6] According to the 2008 proposed nomenclature of DSD, Swyer syndrome falls under the XY umbrella of DSD which encompasses any disorder in which chromosomal, gonadal or anatomic sex development is abnormal. In Swyer syndrome, mutations or deletions occur in SRY region in 10-20% of cases and in 80-90% of patients genes located on autosomes or X chromosomes are affected.[4,10] These mutations are usually denovo, and only few familial cases have been reported. Familial inheritance may be linked to X or Y chromosome and may be autosomal dominant or recessive.[7]

A protein produced due to gene on SRY region causes differentiation of gonads into testes.[8,9] Mutation of this gene leads to defective production of proteins which in turn leads to streak gonads (a form of fibrous tissue) bilaterally during early embryogenesis despite XY karyotype. These streak gonads are non-functional and hence absence of release of testosterone leads to development of female phenotype and external genitalia and hence the sex of rearing is female. Streak gonads are unable to secrete Antimullerian hormone (AMH), so mullerian ducts develop to form uterus and fallopian tubes and hence internal organs are also female. These patients are usually tall statured with eunuchoidal habitus. Streak gonads fail to produce estrogen at the time of puberty and lead to hypoestrogenic state, hence these patients usually present with primary amenorrhoea. Due to normal female external genitalia, sexual function remains normal. Infertility occurs due to absence of ovarian tissue and hypoestrogenemia.[6]

The diagnosis of Swyer syndrome is usually delayed due to the normal female phenotype and normal sexual function as was in our case.[6] As streak gonads fail to produce estrogen, uterus usually remains hypoplastic. Due to hypoestrogenism, HRT is essential for initiating menarche, for breast development, for regularizing menstrual cycle and enabling endometrial preparation for an ART cycle. Pregnancy can be achieved after endometrial preparation in

ART cycle with egg donation.[16-43] Many of these patients do not respond well to HRT probably because of lack of estrogen receptors in the endometrium. None of the published articles mention the time taken for endometrial preparation prior to ART. However once the pregnancy is achieved, the presence of XY genotype and the H-Y antigen does not affect the pregnancy growth and the uterus is able to sustain the pregnancy successfully. However, these pregnancies are at an increased risk of obstetric complications, like gestational hypertension, pre-eclampsia, preterm labour and fetal growth restriction (FGR).[11,12]

According to the review of literature out of the 29 patients of Swyer syndrome (excluding our case) who achieved a successful pregnancy, 1 patients were lost to follow up.[21] Out of the remaining 28 patients there were three patients who delivered twice.[22,23,43] and Michala et al reported three deliveries in the same patient of Swyer syndrome.[6,28] We present the 30th case of swyer syndrome patient who delivered twins at our centre.

Table 1 depicts an updated list of all the 29 patients of Swyer syndrome with successful pregnancy and their obstetric outcome. (Table 2, Fig 1).

Most of the swyer syndrome patients had a singleton pregnancy. Only 10 twin deliveries have been reported in world literature. We report the 11th case of twin delivery in Swyer syndrome. Most of the deliveries were caesarean sections (85%) for various indications like gestational hypertension pre-eclampsia, android pelvis, failed induction, twins, malpresentation of twins, FGR, fetal distress, breech, premature rupture of membranes (PROM). In our case, the indication of caesarean section was ICSI conceived twins with GH and FGR. Only 5 patients (15%) delivered vaginally.[15,16-43] Some of the neonates required care in neonatal intensive care unit (NICU) because of prematurity or FGR. Interestingly, most of the patients of Swyer syndrome breast fed their babies as lactation is dependent on an intact hypothalamic pituitary axis which is common to both men & women.

Patients with Swyer syndrome have an increased risk of malignancy in gonads (gonadoblastoma), which could be a precursor of dysgerminoma. The risk is estimated at about 15-30%.[8] Sometimes these gonadal tumours can develop at a younger age even before the diagnosis of Swyer syndrome is suspected. However early diagnosis is important because prophylactic gonadectomy of dysgenetic gonads is recommended to prevent malignancy. Other risks of gonadal dysgenesis include prolonged hypoestrogenemia with osteoporosis and

virilisation.[9,13,14] Hence they should be started on HRT after lactation to protect their bones from osteoporosis.

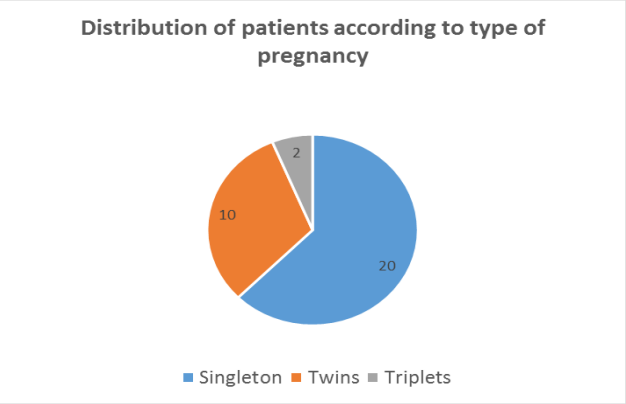
Table 1: Reported pregnancies in patients with Swyer Syndrome and Obstetrical outcome.

S. No.	Authors	No. of attempts	Age at delivery (years)	Pregnancy single/twin /Triplet	Complications	Weeks at delivery	Mode of delivery with indication
1	Lutjen P.[16] 1984	1	25	Single	None	38	CS (ART Pregnancy)
2	Frydman et al[17] 1988	1	37	Single	None	41	CS (ART)
3	Sauer et al[18]1989	1	28	Twin	Preeclampsia (PE)	35	CS: Severe PE
4	Coenet et al[19] 1990	1	25	Single	None	36-41	CS: Android pelvis
5	Bardeguez et al[20] 1990	1	Unk	Triplet	None	Unk	CS: Preterm, PROM
6	Bianco et al[21]1992	1	Unk	Single	None	Unk	Unk
7	Kan et al[22] Oskarsson 1997	2	2 attempts 1st -30 2nd 32	1st Single 2nd Twin	PE	1st >37 2nd 36	1st vaginal delivery 2nd -CS Failed induction with twin with PE
8	Dirnfeld et al[23] 2000	2	30 (1st attempt) 32 (2nd attempt)	1st -Single 2nd -Twin	Spontaneous demise of one fetus at 19 weeks of pregnancy 1st attempt	1st -41 2nd –37	1.CS : Failed induction 2. CS: Failed induction with previous CS
9	Selvaraj et al[24] 2002	1	27	Single	None	Unk	CS: failed induction
10	Chen et al[25] 2005	1	31	Twin	None	36	CS:Malpresentation of twins
11	Ko et al[26] 2007	1	36	Triplet (twins alive & H mole in one)	PE	33	CS: PE and proteinuria
12	Plante and Fritz[27] 2008	1	27	Single	None	38	CS: Non descent of head
13	Michala et al[28] 2008	3	Unk	Single	1-none 2-none 3-PE	1&2-unk 3rd - 36	1-NVD 2-NVD 3-CS-Preeclampsia
14	George Creatsas et al[29] 2011	1	35	Single	Preexisting hypertension	Unk	CS: FGR & Fetal distress

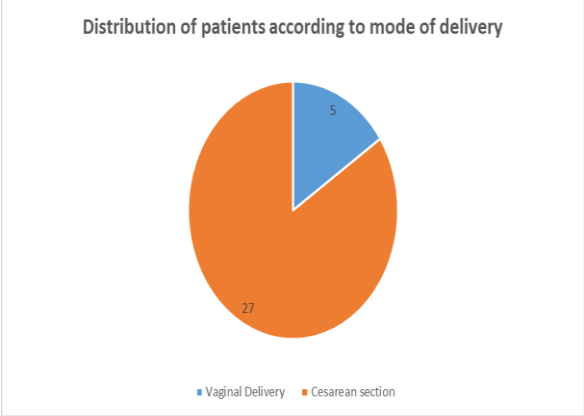
15	Tulic et al[30] 2011	1	30	Single	Oligohydramnios	39	CS: Breech with oligohydramnios
16	Gao et al[31] 2011	1	32	Twins	None	Unknown	CS (Twins)
17	De Santis et al[32]) 2013	1	27	Twins	None	35	CS (Twins)
18	Murtinger et al[33] 2013	1	30	Twins	None	36	CS (Twins)
19	Taneja et al[34] (2016)	1	36	Twins	Vanishing twin at 6 wks Single pregnancy	39	CS: Single
20	Aradhana kalra et al[35] 2016	1	27	Single	Severe PE with morbid obesity	34	CS: FGR, PE
21	Rekha Rajendra kumar et al[36] (2017)	1	27	Single	None	36	CS PROM
22	Shah et al[37] 2018	1	32	Twins	PIH	34	CS Twins with PE
23	Andreas chrysostomou et al[38] 2019	1	24	Single	None	38	CS
24	Gupta A et al[39] (2019)	1	1st sister-25 yrs	Single	None	37	NVD
25	Gupta A et al[39] (2019)	1	2nd sister 27 yrs	single	GH	37	CS
26	Izabela Winkler[40] 2022	1	Unk	Twins	Unidentified problems	33	CS: FGR in one fetus with risk of asphyxia
27	Siddique et al[41] 2008	1	Unk	Single	None	38	Vaginal delivery
28	Azamsadat et al[42]	1	Unk	Single	preeclampsia	37	CS PE
29	Urban et al[43]	2	32 (1st attempt), 34: (2nd attempt)	1st –Single 2nd -Single	No evidence	1st- 40 2nd 39	CS CS
30	Jindal et al (Our case report)	1	32	Twins	Oligoamnios, FGR, GH	37	CS: Twins Oligoamnios, FGR & GH

Table 4: Distribution of patients according to indication for caesarean section.

Type of pregnancy	No. of pregnancies
Singleton	20 (63 %)
Twins	10 (31 %)
Triplets	2 (6 %)



Mode of delivery	Number of cases
Vaginal birth	5 (16%)
Caesarean Section	27 (84%)



Indication for cesarean	No. of Cases
ART	12
Preeclampsia	5
Android pelvis	1
Preterm labor/ PROM	2
Failed Induction	4
Malpresentation	2
FGR with fetal distress	4
Non descent of fetal head	1
Unknown	1

CONCLUSION

Diagnosis of Swyer syndrome is often delayed due to female phenotype and normal sexual function. Treatment of Swyer syndrome requires a multidisciplinary approach. Prophylactic gonadectomy for prevention of malignancy, hormone replacement therapy for induction of puberty, treatment of osteoporosis and psychological support are important aspects of patient's care. Achieving a pregnancy in Swyer syndrome (XY) with ART with ovum

donation cycle is a rare feat and highlights the nature's way of procreating even in a genetic male against all odds.

We need more such case reports on pregnancy outcome in patients with Swyer syndrome to expand our knowledge and better define the management of this unique patient population.

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