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Twin Discordance Secondary to Suspected Spontaneous Superfetation: A Case Report.

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Abstract

Spontaneous conception of an additional fetus in the presence of an ongoing pregnancy is quite rare and remains a puzzle in the human reproductive cycle. Although several theories have been proposed, none has satisfactorily explained the mechanism of this deviation from the natural human reproductive events. We report a case of a set of twin with dichorionic placentation, discordant birth weight, maturation and estimated Gestational Age (to the tune of 4 weeks) necessitating variation in their neonatal care plan. Mother did not employ any form of Assisted Reproductive Technique (ART) in their conception. The authors diagnosed possible twin superfetation. Its rarity in human prompted this report as an addition to the few existing reports and also to raise a higher index of suspicion among clinicians concerned with the delivery and care of newborns.

Key words: Twin, discordance, superfetation, conception, gestational age

INTRODUCTION

In superfetation, there is conception of an additional fetus during an ongoing pregnancy. This occurs at an interval as long as, or longer than an ovulatory cycle resulting in a situation where babies of different gestational age co-exist during pregnancy (Gary et al. 2001). Though a feature in animals, it is rare in human race that only fewer than ten cases have been reported in medical literatures. (https://www.researchgate.net/.../322275983)_ Superfetation_in_Humans_-_Myth_or_Real).

In Humans, a complex interplay of hormonal activity ensures mono-ovulation per cycle and suppression of further cycle and ovulation in the presence of a successful implantation and developing fetus. Although the pathophysiology of superfetation in humans is not fully understood, several theories are in existence, one of which proposed that the discordance in the gestational age of coexisting twins could be due to a Luteal-Out-Of-Phase (LOOP) like events which occur between1-3weeks after the ovulation that caused the first pregnancy (Juan et al. 2012). The LOOP like event results in atypical increase in E_2 in the mid-luteal phase which may allow passive sperm transport from the vaginal fornix to the fallopian tube ipsilateral to the ovulatory ovary and trigger a LH peak and ovulation. Also in the very early age of pregnancy when the decidua reaction is still incomplete and at-least one proximal fallopian ostium still open, spermatozoa can ascend to the fertilization site and the extra fertilized embryo(s) descend to the implantation site(s). Furthermore, with the advent of Assisted Reproductive Techniques (ART), it has been recognized that natural barriers to superfetation can be overcome. Cases of superfetation resulting from ART with or without additional spontaneous conceptions have been reported (Amsalem et al. 2001; Harrison et al. 2005). A case of artificially induced superfetation had also been reported by Dmowski (Dmowski et al.

1997) over a decade ago. What may appear like a variant of superfetation had been reported in a gravida 4 woman with double uterus by Ibrahim et al (2009). On the strength of the supporting evidence of superfetation and rarity of such cases we make a report of babies IC 1 & 2 suspected to be a case of twin superfetation. It is expected that this report will add to existing incidences and raise a higher index of suspicion among Obstetricians and Neonatologist when managing discordant twins.

CASE REPORT

Babies IC 1 & 2 are a set of twin delivered via Emergency Cesarean Section (EMCS) in our center by a 23year old un-booked mother. Indication for the EMCS was severe preeclampsia and a previous uterine scar and mother was not sure of her date. She had Ante-Natal Care (ANC) at a private hospital and said to have received 1 dose of Tetanus toxoid but no antimalarial Intermittent Preventive Therapy (IPT). An Ultrasound done at 24weeks revealed a singleton viable fetus, no coexisting uterine mass or any structural uterine defect was reported. No history of use of any form of Assisted Reproductive Technique (ART) in the index or previous pregnancies. The pregnancy was normal until a day before delivery when her blood pressure was noted to be too high during ANC visit in the private hospital necessitating emergency referral to our center for further cares. She was admitted and had an EMCS. Intra-operatively, dichorionic placentation was discovered and babies IC 1 & 2 were extracted, both cried at birth. The relevant clinical findings were tabulated below.

Clinical findings	**Baby IC -1**	**Baby IC- 2**	
Sex	Female	Female	
Colour	Plethoric	Plethoric	
Jaundice	Nil	Nil	
Temperature	35.0	<35.0	
Heart Rate	160 beats/min	172beats/min	
Respiratory Rate	60 breaths/min	88breaths/min	
Birth weight (kg)	1.9	0.8	
Length	43cm	30cm	
OFC	33cm	25cm	
RBG	3.9mmol/l	1.9mmol/l	
Haemoglobin level (g/dl)	18.7	17.3	1.4
Total SB (direct)	61.5 (9.5) µmol/l	163.3 (27.3) µmol/l	
Blood group	O-positive	O-positive	
MATURITY RATING USING NEW BALLARD SCORING SYSTEM (NBS)			
	Baby IC -1 scores	**Baby IC- 2 scores**	
Skin	1	1	
Lanugo	2	0	
Plantar surface	4	4	
Breast	2	1	
Eye/ Ear	2	2	
Genitalia	2	2	
Posture	4	3	
Square window	4	1	
Arm recoil	4	4	
Popliteal angle	3	2	
Scarf sign	2	2	
Heel	2	1	
TOTAL SCORE	**32**	**23**	
Estimated GA	**36-38 weeks**	**32-34 weeks**	**4 weeks discordant**

Fig. 1: Baby IC-1

Fig 2: Baby IC-2

Mother is 23year old while the father is 32year old. Their blood groups were both O-positive. A diagnosis of Discordant Twins possibly due to spontaneous superfetation was made. Twin IC-1 was diagnosed Low Birth weight with Moderate–Late Prematurity while Twin IC-2 was diagnosed Extreme Low Birth Weight, Severe Prematurity and hypoglycemia. Euglycemia was restored in the latter twin with 2ml/kg of 10% dextrose water and maintained with glucose infusion at 8g/kg/min. Further care of Twin IC-2 was continued in an infant incubator while Twin IC-1 was discharged to the mother after stabilization.

DISCUSSION

Superfetation is quite rare in human. When a suspicion is made there is need to rule out presence of any factor that could have impaired the growth of the smaller twin. Such factors include but not limited to presence of structural uterine anomaly, co-existing intrauterine mass, presence of congenital chromosomal anomaly, twin- twin transfusion syndrome etc. Ultrasounds scan at 24weeks which although failed to detect the presence of twin gestation, however reported absence of any structural uterine anomaly or co-existing mass. The inability of the scan to detect twin at 24 weeks gestation maybe blamed on possible limited sonographic skill or the sensitivity of the equipment. Moreover, it was done in private center where the expertise of the sonographer could not be ascertained. It would have been ideal to have an earlier scan and indeed serial scans to monitor growth progress of the twins as done by Tuppen (Tuppen et al. 1999) who reported a case of spontaneous superfetation in the first trimester and had serial scan for growth monitoring through the pregnancy. In our case our client presented later in the pregnancy and an apparently normal scan report at 24weeks gestation would have in no way be a stimulant for further scans prior to presentation. There were no structural or dimorphic features noted in any of our babies to account for the obvious disparity in growth and maturity. Again subjecting them to extra uterine scans including echo-cardiograph and chromosomal analysis would have been ideal to rule out presence of chromosomal

anomaly. However, these investigations are not without cost and medical bills are paid out of pocket in our setting (Hoare, 1987). Therefore, our clients could not afford them especially in this situation where no anomaly was detected on physical examination. The discordance in the Haemoglobin level of our babies (1.4g/dl) and the plethoric appearance of both twins were not supportive of twin-twin transfusion syndrome (TTTS). The neonatal criteria for diagnosis of twin to twin transfusion syndrome stipulates a haemoglobin difference of 5g/dl with plethoric appearance of the recipient and anaemic appearance of the donor twin (https://emedicine.medscape.com/article/271752-overview).

Hence, the observed difference in the growth, maturity and care need, the difference in gestational ages (4weeks by NBS) and the presence of a dichorionic placentation at birth laid strong credence to the suspicion of twin superfetation. Noting its rarity in humans, the authors believe that reporting this case will add to the existing literatures and stimulate a higher index of suspicion among clinicians caring for the babies at birth.

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