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A Case of Prolonged Pregnancy and a Review of the Literature on Superfoctation in an Endeavour to Explain the Phenomenon

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INTRODUCTION

By superfoctation is meant the fertilisation of two ova during different intermenstrual periods; that is to say, the second ovum is fertilised after the first which was set free from a previous ovulation, and has been developing for a month or more. This must be admitted as a possible occurrence in the first two or three months.

The belief in the occurrence of this phenomenon is of ancient origin. According to Radash its possibility was discussed by Hippocrates, Pliny and Aristotle (Studdiford, 1936). De Lee (1933) states that superfoctation is held possible by American and French authorities, but denied by the British and German.

A survey of the literature describes several varieties of cases which were believed to support this view:—

- Abortions in which two or more foetuses are passed with a marked difference in size and appearance.
- Cases of pregnancy which go to term and deliver two or more foetuses with a marked variation in size, e.g., in bicornuate uterus.
- 3. Cases occurring in animals (Snyder and Wiselocke, 1933). Studdiford's conclusions from the literature are that no proved case of superfoctation in the human being could be found, but that a few cases were found (Longmore and Schaab, 1862) that suggests this phenomena is possible. In order to prove such a case, certain criteria must be fulfilled.

Studdiford described two cases which superficially suggest superfoctation:—

1. The retention of a dead twin, the other twin remaining alive.

2. The prolongation of pregnancy which suggests the possibility of superfoctation, but which leaves no opportunity to submit this explanation to proof.

Gaupin (1951) also describes a case where the second fertilisation took place when the original conceptus was about three months' duration—a six weeks foetus and a foetus of four mouths presented for delivery.

Professor Johnstone considers superfoetation an extremely unlikely occurrence, but he admits that the cases most difficult to explain on any theory are those where the birth of a fully developed child is followed about four months later by the birth of another fully developed child and no trace of a bicornuate uterus can be found. Mayes (1950) is of the opinion that if the case is analysed carefully the explanation will be found to be a bicornuate uterus or merely twins of markedly different sizes due to different rates of development.

Finally, a study of the literature on the duration of pregnancy shows that the duration of pregnancy extends from the moment of fertilisation to the birth of the baby, a condition known as the "fertilisation-labour interval." accepted length of pregnancy is commonly regarded as 280 days, 40 weeks or 10 lunar months. Fully developed children have been recorded as being born after gestations as short as 240 days and as long as 313, 320 and even 331 days from the commencement of the last period. The longest period of gestation which has ever been admitted in the law courts in Great Britain is 331 days, but the judgment was based on negative findings rather than positive evidence (Gaskell v. Gaskell; Times Law Report, 21st September, 1931).

The case reported was considered worthy of publication, particularly by reason of a possible superfoctation, a lengthy pregnancy of 383 days' duration from the last menstrual period to confinement, terminating in a dramatic fashion. The case was remarkable in that various complications and a variety of intercurrent illnesses occurred during the course of the pregnancy.

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CASE REPORT

The patient, a healthy European woman of 35 years of age, was a multipara 2, with two healthy children, a son aged seven years and a daughter aged five years. There was a history of an abortion at three months between the first and second pregnancies. She had a cholecystectomy performed in Cape Town in 1952.

Obstetrical History.—The patient lived in Johannesburg prior to coming to Salisbury four years ago. Her pregnancies and confinements and post-natal history were normal. Both her children were breast-fed. The author attended the family for some three years before the patient consulted him on:—

22.10.53 because of a possible early pregnancy. She stated that her last normal menses was on 6th August, 1953. Since the birth of her second child her menstrual history had been regular—habit 28 days, type 5-7 days. She admitted to the likely possibility of pregnancy, as she and her husband, though practising contraceptive measures at times, were happy at the prospect of an addition to the family. (77 days since L.M.P.)

Clinical examination revealed a healthy woman-height 5 ft. 10 in., weight 158 lbs. All systems were normal and there were early signs of pregnancy—breast changes and an enlargement of the uterus. A presumptive diagnosis of pregnancy was made, the findings corresponding to a ten weeks gestation.

26.10.53: The patient was seen at her residence in Borrowdale. There was a history of sudden onset of bilious vomiting for some hours and the appearance of a slight P.V. loss that morning. There was no backache or pelvic pain consistent with an impending abortion; there was no passage of clots. There was a fever of 99.4° F, and a pulse rate of 84. Within 24 hours the slight haemorrhage had ceased, but the nausea and vomiting persisted for a further two days. (81 days since L.M.P.)

The prodromal stage of infective hepatitis was suspected, but no sign of icterus, enlargement of the liver or spleen appeared. Urine examination was also negative for bilirubin and bile slats.

12.11.53: The patient was visited owing to a febrile illness, pyrexia, aches and pains and general malaise. Temperature 100° F., pulse rate of 90. Examination revealed the uterus corresponding in size to a 12 weeks gestation. Breast signs were still present and breast activity admitted. The febrile illness appeared to be a "virus influenza." Blood smears to exclude malaria parasites were taken and found negative. Treatment was symptomatic. The temperature and all symptoms abated by the third day. (98 days since L.M.P.)

18.11.53: The patient reported for a full ante-natal examination. She complained of extreme weakness, lassitude and vertigo with morning sickness and constipation. There were subjective symptoms of a post-influenzal debility with pregnancy and mild hyperemesis. Clinical examination confirmed a diagnosis of pregnancy.

The uterus was enlarged, the fundus being three fingers above the symphysis pubis. It was soft, mobile and anteverted. The B.P. was 118/70 and Hb. 71 per cent., and the urine examination negative. The weight was 162 lb. Cytacon, 50 mg. injections three times weekly were commenced, as well as hexadoxin tablets and an iron tonic after meals. (104 days since L.M.P.)

4.12.53: The patient reported that she had a complete cessation of symptoms of pregnancy for the last three days, with absence of nausea, frequency of micturition and no breast symptoms. She was feeling fitter than

for some weeks past. On examination her colour was good, B.P. 118/70. The weight dropped to 159 lb. The uterus was not palpable on abdominal examination. P.V. examination—uterus retroverted and in size smaller than on the examination on 18.11.53, i.e., 16 days previously. On questioning, the patient assured me she had had no further P.V. loss and no lower abdominal pains or backache. (120 days since L.M.P.)

I had anticipated finding a uterus corresponding in size to that of an 18 weeks gestation, but instead it was one of only 10 weeks' duration. Quickening had also been expected from the fourth month onwards, but there were certainly no symptoms of this.

At this juncture one had to consider the possibility of a missed abortion, the pregnancy having been interrupted by the febrile illness of 12.11.53.

The patient was informed that though the signs of pregnancy still persisted in some degree, it would be necessary to wait four weeks for a further examination before any further active measures could be taken. This advice did not entirely satisfy the patient, who subsequently became very emotionally upset.

7.1.54: The patient reported at my consulting rooms in good form. She had experienced a return of symptoms of pregnancy (breasts, bladder, etc.). On examination her weight was $163\frac{1}{2}$ lb., a gain of $5\frac{1}{2}$ lb. in four weeks. The uterus was only of a size corresponding to a 12 weeks gestation, and on this finding and a suspicion of the patient having retained an abortion which might be taking on the changes of a carneous mole, I felt justified in putting her on a 14-day course (or until menstrual bleeding recurred) of Esthynl, 0.05 mg. tablet daily. (154 days since L.M.P.)

15.1.54: The patient reported and complained of persistent nausea, but no vaginal loss. A catheter specimen of urine was taken and this was submitted to a private laboratory for a xenopus test, which was reported as being positive. (162 days since L.M.P.)

In view of these findings, the patient was advised to seek a specialist's opinion and she was examined on 15.1.54 and again on 22.1.54 by a gynaecologist, who reported, "An enlarged soft uterus, corresponding to a 12 weeks' pregnancy." The patient and her husband accepted this reluctantly, but to their urgent appeal for a curettage to be performed for fear of a deformed child and other future complications, medical opinion was in favour of "masterly inactivity," that cardinal rule in midwifery. I am convinced the patient was on the point of "ringing the changes" and seeking more sympathetic advice from another of my colleagues, but by gentle persuasion, sedation and morale boosting, the patient carried on in her travail until her next visit on:

12.2.54: She looked fit. There were no toxic symptoms. B.P. readings were low, 118/70. Urine examination—N.A.D. The uterus was enlarged and soft, corresponding to a 14 weeks gestation, i.e., approximately 91 days gestation. Her weight was now 164 lb. (190 days since L.M.P.)

5.3.54: Antenatal visit revealed an enlarging uterus. (211 days since L.M.P.)

29.3.54: Antenatal visit. The fundus had reached the level of two fingers breadth below the umbilicus, but still no foetal movement had been experienced by the patient. Her weight on 5.3.54 was 166 lb. (235 days since L.M.P.)

There were mild symptoms of a left sided pyelitis with pus cells in the urine (29.3.54). The patient responded rapidly to treatment with fluids, sulphatriad tablets and mist, pot, cit. Her weight was now 168½ lb.

Arrangements were made at this visit for:-

- (1) A repeat xenopus pregnancy test.
- (2) A straight X-ray of abdomen.
- (3) Haemoglobin, antibody and sensitivity tests, in view of the patient's known blood group—"O," Rh, negative.
- 6.4.54: Reports received on the above tests:-
 - (1) The xenopus pregnancy test was negative.
 - (2) X-ray of the abdomen: "Uterus enlarged about three fingers breadth above crest of ileum. The foetal parts are demonstrated in the prone position. Foetal spine is straight and this suggests it is alive. The size of the foetal parts, however, is not large enough for 20 weeks.
 - (3) Hg. 91 per cent. Abnormal antibodies—nil. Sensitisation tests—negative.
- 15.4.54: Patient 'phoned to say that in the last 10 days she had lost 4 lb. in weight to 164 lb. and that she still had not felt the foetus move.
- 17.4.54: The patient was confined to bed suffering from a sudden acute attack of gastro-enteritis. A visit was made and her condition was satisfactory. There was no previous history of amoebic dysentery, but in 1952 the patient had a cholecystectomy performed at Cape Town for gallstones. Temperature 99.4° F., pulse rate 88. There was no dehydration and abdomen was flaccid. The uterus was palpable, the fundus being at the level of the umbilicus. The patient responded rapidly to sulphaguanidine suspension, fluids and diet. (254 days since L.M.P.)
- 28.4.54: Antenatal visits—feeling well and she had been conscious of vague movements in the uterus. Weight was now 170 lb. Blood pressure readings and urine satisfactory. Abdomen—fundus of uterus two fingers breadth above the umbilicus. The uterine souffle was audible for the first time. (265 days since L.M.P.)
- 3.5.54: X-ray of abdomen repeated. Report—"Film now shows definite viable foetus corresponding to a 20 weeks gestation. (279 days since L.M.P.)
- 14.5.54: Recurrence of attack of gastro-enteritis—mild and no pyrexia. Had two mild attacks of syncope. Confined to bed for 48 hours.
- 15.5.54: Husband's serological tests repeated. Report—Group "A," Rh. positive, probably homozygous.
- 31.5.54: Antenatal examination—patient feeling very well. Weight now 176 lb. Foetal movements active for past three weeks. Examination of abdomen—the fundus of uterus midway between the umbilicus and the xyphisternum. E.D.D. (estimated from second X-ray findings on 3.5.54) corresponding to 30 weeks. (298 days since L.M.P.)
- 26.6.54: Patient had a third attack of gastro-enteritis. Responded to treatment and symptom-free in three days. (324 days since L.M.P.)
- 14.7.54: Antenatal examination. Patient is feeling and looks fit. All systems normal. B.P. 118/70. Urine examination—N.A.D. Abdomen—uterus corresponds to 32 weeks gestation. Foetal movements strong. Foetal heart heard. Now six weeks from term, i.e., 30 weeks or 210 days. Weight, 178 lb. (343 days since L.M.P.)
- 20.7.54: Visit—patient suffering from a typical rubella infection, with rash, pyrexia and cervical adenitis and enlarged post auricular glands. Illness lasted three days to 23.7.54. (348 days since L.M.P.)

- 20.8.54: Antenatal examination. Patient reports that foetal movements have been quiet during last week. Examination—weight, 182 lb. Abdomen—foetal position, breech in R.S.A. position. External version attempted and failed. (379 days since L.M.P.)
- 24.8.54: Abdomen X-rayed. Report as follows: "Single foetus L.S.A. position with extended legs."

Patient was admitted to the Lady Chancellor Maternity Hospital in forenoon. At 3 p.m., under sodium pentothal and gas and oxygen, further attempt at internal version proved unsuccessful. Medical induction with prostigmin two hourly, commenced at 4 p.m., 2 x 15 mg. tablets. At 6 p.m. the patient's condition was satisfactory. Complained of a few vague abdominal pains. Prostigmin 1 x 15 mg. tablet given. At 7 p.m. a sudden brisk haemorrhage with clots P.V., followed by acute pain in left side of uterine wall. Tonic contractions suddenly stronger. At 7.15 p.m. I examined the patient. Her condition was good and no symptoms of shock. Pulse rate 80, B.P. 120/74. Abdomen acutely tender on palpation over fundus and to left side of uterus. P.V. examination—"multip" os of one finger dilatation. Placenta felt like a sponge. Diagnosis—a central placenta praevia. The patient was given morphine, gr. 4, and arrangements for an immediate caesarian section made. (383 days since L.M.P.)

At 7.45 p.m., under sodium pentothal with ether and gas and oxygen anaesthetic, a classical caesarian section was performed. The uterine vessels all over the lower segment were greatly distended. A viable female child was extracted. Haemorrhage was free. The placenta was firmly attached centrally on the lower segment and had to be stripped off. The operation was completed by 8.40 p.m. Condition of both mother and child was satisfactory. Placenta was of average size—appeared healthy apart from small infarcts present in the substance. The membranes were apparently intact.

Baby was small, but of good colour; the nails were well formed, but there was little subcutaneous fat.

25.8.54: The patient's condition was good. Three pints of group "O," Rh. negative blood was given. A Hb. estimation taken before the transfusion was 58 per cent. The baby's condition was fair. She appeared lethargic. Laboratory report on blood taken from baby 28.8.54: Hb. 106 per cent. = 18.24 per cent. (100 per cent. = 16 mg. per cent.). No sign of antibodies in child's blood, moderate polychomasia. Direct Coombes test was negative.

Weight Recordings During the Pregnancies

4
4
3
4
l
$4\frac{1}{2}$
4
$5\frac{1}{2}$
6
2
2

These weights were recorded on my consulting room scale on the specified days. The patient gained 22 lb, in weight during her pregnancy.

27.8.54

1.9.54

Blood Pressure Readings

					0		
(first	visit	:)		*****	*****	100/70	
		*****		*****	******	118/70	
*****		*****	*****	*****		120/74	
*****						128/80	
		*****			*****	130/80	
*****	*****	******				118/70	
*****	*****	*****	******	*****	*****	120/74	
Haemoglobin							
*****		*****	*****	*****	81 p	er cent.	
		*****	*****		71	**	
	*****				73	**	
*****					91	-11	
*****	******	gg++++	*****	*****	80	77	
		<i>H</i>	Haemog	Haemoglobin	Haemoglobin	Haemoglobin	

(Three pints of blood given 25.8.54.) Intercurrent Illnesses During the Pregnancy in Chronological Order

58

74

26.10.53: Pyrexia and bilious attack + P.V. loss for 24 hours. Infective hepatitis suspected, but no such developments. ? Second ovulation.

12.11.53: Virus influenza. Brucellosis agglutination reaction negative.

29.3.54: Symptoms of left sided pyelitis (mild),

12.4.54: Acute gastro-enteritis.

14.5.54: Acute gastro-enteritis.

26.6.54: Acute gastro-enteritis.

20.7.54: Rubella (German measles).

Laboratory Reports

Blood groups-

The patient: blood group "O," Rh. negative.

The husband: blood group "A," Rh. positive (probably homozygous).

Conclusion.—No evidence of erythroblastosis foetalis. Subsequent post-natal course was uneventful. Breast feeding was established on fourth day, i.e., 28.8.54, and a further Hb. determination on the mother's blood taken on 29.8.54 was 74 per cent. Mother and child were discharged from the home on 29.8.54.

The history would suggest a very ill woman enduring a 12 months' pregnancy with complications and intercurrent illnesses, while the uncertain termination to the ordeal would promote a progressive mental anxiety and emotional stress. Nevertheless the patient kept wonderfully well between the varied indispositions, and she was able to conduct her duties as a housewife and even entertain and do the social rounds with her husband almost up to the time of her sudden and dramatic confinement.

Discussion

On reviewing the literature on superfoctation one finds that authorities in Britain will not admit definitely to its occurrence (Johnstone). American gynaecologists do, however, consider that the condition may occur. Certain criteria are essential to prove indisputably that cases of this type are in fact superfoctation. Explanations have been given in all cases reviewed in the literature which can conceivably disprove such a phenomenon.

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A bicornuate uterus could prove a case of presumed superfoctation. In this case caesarian section revealed no such abnormality.

Studdiford (1936) does not place much emphasis on the duration of pregnancy as being a supporting factor in cases of superfoetation; in this study one finds in the British literature that the longest period of gestation which has ever been admitted in the law courts in Great Britain is 331 days.

In the case under review the actual length of gestation from L.M.P. to the actual birth was one of 383 days' duration.

The P.V. loss with a pyrexial illness lasting 24 hours, 81 days from the L.M.P. (6.8.53), may in fact have been a second ovulation with a second nidation following this date. This would account for the prolonged antenatal period. As a hypothesis it is conceivable that the first pregnancy terminated at this time with subsequent cessation of symptoms and signs of pregnancy some 119 days from L.M.P., and the reappearance of these phenomena of pregnancy 152 days from the last menstrual period.

The weight checks also conformed with the patient's subjective observations and my own clinical findings at this stage of the pregnancy.

The delayed "quickening" is difficult to explain apart from the fact that it was apparent that the foetus was extremely small and inactive at the time of the first radiological examination on 29.3.54, i.e., 233 days from the last normal menses. The patient was definitely aware of "quickening" for the first time on 28.4.54, a matter of 274 days from L.M.P. and 166 days from the assumed second ovulation and second nidation.

In view of the Rh. factor, the patient being blood group "O," and Rh. negative and her husband blood group "A" and Rh. positive (probably homozygous), and a history of one abortion at three months after the second child of five years, the possibility of Rh. incompatability and antibody formation being concerned in the case was a constant feature, but one which never appeared in the various serological tests during the pregnancy.

The estimated date of delivery from L.M.P. on 6.8.53 was 13th May, 1954; the actual confinement took place on 24th August, 1954, i.e., 103 days later.

Accepting the possibility of a second ovulation on 26th October, 1953, i.e., 81 days from L.M.P., and a second fertilisation occurring within seven days of that date, then the estimated date of delivery of the second foetus was 2nd August, 1954. The confinement took place 22 days later, i.e., 24th August, 1954.

The baby's birth weight, 5 lb. 12 oz., was well below average for term; the reason for this may have been related to some metabolic factor, but certainly not to any apparent toxaemic factor, as the case was unique in that no signs of toxaemia of pregnancy occurred.

The placenta and membrane were only examined macroscopically. No microscopic examination being conducted eliminated any factual proof and evidence of a possible second gestation in the nature of a carneous mole or mummified area in the placenta area. This omission put the case without the necessary criteria in a case of superfoetation.

A case of possible superfoctation has been described and discussed. In view of the size of the foctus presenting at birth, 5 lb. 12 oz., the second ovulation on 26th October, 1953, and a second pregnancy seemed the more likely explanation rather than a baby of that weight arriving 381 days after the L.M.P., taking into account any metabolic factor which may have retarded the baby's weight.

This case then conforms to the one of two reported by Studdiford, where there was a prolongation of pregnancy, which suggests the possibility of superfoctation, but which left no opportunity to submit this explanation to proof.

The patient was a highly intelligent woman under constant supervision by her doctor. That meant that there was factual evidence in the case well beyond the bounds of doubt and supposition.

SUMMARY

(1) A case of a European woman, multipara 2, who had a pregnancy lasting 383 days, is presented.

- (2) The history suggested a second ovulation occurring 79 days after the last menstrual period with interruption of the pregnancy at that time; a second nidation occurring within a week of this date.
- (3) The phenomenon caused one to consider the rare case of superfoctation to account for the prolongation of pregnancy.
- (4) The literature on superfoctation has been studied, and criteria to meet the requirements of such a condition have been considered and discussed in relationship to the case.
- (5) It has been found impossible to submit this explanation of superfoctation to proof in the reported case.
- (6) In view of the dramatic conclusion of the pregnancy and a classical caesarian section being performed, it was possible to disprove a condition of bicornuate uterus.

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