DOES SUPERFOETATION OCCUR?**REPORT OF A POSSIBLE CASE**

BY

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Superfoetation may be defined as the implantation of a second fertilized ovum in a uterus which has already contained a pregnancy for one month or more. The possibility of such a phenomenon occurring in the human female is still a matter of controversy, and the following case report, although unfortunately scientifically incomplete, will be of interest in view of its extreme rarity.

Case Report

A woman, aged 35, married for eight years, gave the following amazing obstetrical history. In 1930, the year following her marriage, she was delivered of a stillborn child at thirty-two weeks. In 1931 she had another stillbirth, this time at twenty-eight weeks. In 1932 she was delivered of stillborn twins and a further pair of twins were again stillborn in the following year. In 1934 a third pair of twins, which did not live, were born at about twenty-eight weeks. She did not know the cause of death of any of these babies, but thinks that the first was "trouble with the cord." She also believes that some of them were malformed. There was no family history of twins on either side. In the following year, 1935, she had a miscarriage following an accident at about sixteen weeks. On March 2, 1936, she had another miscarriage, this time at about twelve weeks.

Following this miscarriage she had two small "shows" at intervals of a month, the first two months after her miscarriage. Apart from this, amenorrhoea existed from the miscarriage on March 2 to December 20, 1936, when she gave birth to a normal live male child weighing just under six pounds. The delivery occurred under unusual circumstances. The patient received no ante-natal care, and while motoring in the country experienced three very severe abdominal pains at intervals of one minute; immediately afterwards the child was born in the car, the placenta following almost at once. The husband was the only witness of the delivery and the birth was promptly registered. No doctor attended her. The child survived, and was bottle-fed as the mother had no milk. The patient was little disturbed by the incident, and was about again in twelve days' time. During the three months following the birth she felt perfectly fit and well, but noticed the abdomen remained rather large and was progressively swelling.

She was first seen by us on March 19, 1937, exactly eighty-nine days after the delivery, and said she thought she had developed a cyst. Amenorrhoea had existed since the child was born. No movements had been felt. On examination the breasts were active. The uterus contained a foetus with the vertex presenting in the right occipito-anterior position. The foetal heart was heard. The gestation period was estimated at thirty-four weeks, and there was no apparent abnormality. The Wassermann reaction proved negative.

When told that she was pregnant she flatly refused to believe it, and partly to convince her a radiograph was taken. The radiologist confirmed the diagnosis and position of the foetus, but estimated the period of gestation at thirty-six weeks. Even when shown the film she would not believe she had a normal pregnancy, and was convinced she would produce a monstrosity of some type.

Three days later labour began with spontaneous rupture of the membranes. Progress was normal at first, dilatation of the os being very rapid. After a two-hour second stage with weak pains delivery was terminated by a low forceps operation, as a secondary uterine inertia was feared. While per-

forming the delivery it was noted that the soft tissues stretched very easily. A living female child was secured weighing 6 lb. 12 oz. and appearing perfectly normal and at full term. The third stage was completed within five minutes. Neither placenta nor membranes presented any abnormality, but it was noted that the placenta was rather small, measuring four inches in diameter and three-quarters of an inch in thickness. Bimanual examination fourteen days later revealed no clinical evidence of double uterus, and the patient would not submit to lipiodol injection. Both mother and child made an uneventful recovery, milk appearing in the breasts normally on the third day. At the time of writing both her children are well and gaining weight rapidly.

Possible Explanations

There are three possible explanations for such a case. In the first place the delivery of a live child in December, 1936, may have been a product of the patient's imagination. Against this we have the husband's confirmation, the inspection of the child, of the birth certificate, and of the form of registration by one of us (F. L. McL.), and the rather fragile direct evidence of a small placenta and very easily dilatable maternal passages at the second delivery. On this and the knowledge that several similar cases are to be found in the older literature we feel convinced that the case is genuine. A second possibility is that the case is one of twin pregnancy with retarded development followed by delayed delivery of one of the foetuses due to diminished blood supply and difference in growth rate. The patient's tendency to twin pregnancies perhaps favours this explanation, but it seems hardly credible that the second twin remained for twelve months in the uterus to be delivered alive and apparently perfectly normal. Nearly all such cases end in early abortion. The third explanation is that this is a case of true superfoetation. This will explain the facts of the case, and the times at which we know coitus took place make possible the second fertilization some three months after the first.

Commentary

Theoretically fertilization of another ovum should be possible providing that the cavity of the uterus is not already closed by the developing ovum and that ovulation has continued during the pregnancy. As regards the first condition, we know that the uterine cavity normally becomes completely filled by the developing ovum between the third and fourth months, so that up to this time a second fertilization should be possible. The blockage of the cervix by a mucus plug and the sealing off of the Fallopian tubes by a decidual mucosa at this time have been cited as further objections. Actually the formation of a mucus plug is not by any means constant in the pregnant uterus, and if present it is not always an obstacle to the advance of sperm. The mucus plug has also been noted in the non-pregnant uterus, and it is possible that the ejection of semen from the male organ may be strong enough to pass it or wash it away. In addition, cases in which the menstrual cycle continues during pregnancy indicate an open passage. A final point is that cases have been noted in which the orifices of the tubes were patent in the third month. One may presume, therefore, that though the passage for sperm up the genital tract is closed in most cases of early pregnancy, there are occasional cases in which the passage remains patent up to about the third month.

It is generally believed that ovulation ceases during pregnancy under the inhibitory influence of the corpus luteum. There is evidence, however, that in some cases ovulation does take place. Ravano (1907) examined the

ovaries of 100 pregnant women. He often observed ripening follicles, and saw in no less than 5 per cent. a recently ruptured corpus luteum. Studdiford (1936) examined the ovaries taken from 100 cases of ectopic gestation, and concluded that though in some cases maturation of the ovum took place during pregnancy it usually terminated in degenerative changes and atresia. Moench (1927) reports a case of double uterus in which a miscarriage at twenty-eight weeks was followed three days later by the abortion of a twelve-weeks foetus. Here ovulation must have continued or begun again about the fourth month. Radasch (1921) quoting Cosentino describes the case of a woman who died in the sixth month of pregnancy; the ovary showed a recent corpus luteum, maturing follicles were present in various stages of development, and one ruptured follicle was discovered which still contained a ripe ovum. It seems likely that, in an organ in which functional pathological changes are so commonly met with, the inhibiting corpus luteum influence may occasionally be suppressed and ovulation occur. In cases of repeated abortion the influence of the corpus luteum is thought to wane between the third and fourth months. It is possible that ovulation can take place in these cases at this time, and that if the passage to the ovum is clear a second fertilization might ensue. It is interesting to note that the case we describe had two shows of blood at intervals of a month during the early gestation period.

Similar Cases

We cannot find any cases similar to the one we report in the recent literature, but several such cases are referred to by the older writers. The explanation may be that in modern obstetrics, in the normal course of events, the double pregnancy would always be recognized at the time of delivery and any delay of the second birth dealt with. Twins often vary in size and development, and Schwaab (1920) describes a case of twins weighing 1,900 and 2,850 grammes. It is possible that such cases may be ascribed to superfoetation, and if left to nature the smaller foetus might remain in the uterus till term. A similar case to ours is referred to by Milne (1871). This woman delivered a full-term infant on November 17, 1807; a second infant, also at term, was delivered on February 2, 1808. Churchill (1846) quotes the case of a woman delivered of a full-term child on April 30. On September 17 she produced a second child, which also appeared to be at full term, and both survived. That the uterus was single was confirmed at post-mortem when this woman died in later years.

Other such cases are referred to by Leishman (1873), Barnes (1884), Cassan (1826), and Arrowsmith (1834). Bonnar, in 1864, published a paper describing an investigation of the records of the British peerage; here the exact date of birth of successive children of peers is given without reasonable possibility of error. He has collected several examples of births rapidly succeeding each other which seem inexplicable by any theory other than superfoetation. He cites one case of a child born on September 12, 1849, to be followed by another on January 24, 1850, an interval of 127 days; both children survived. Such cases, he holds, are examples of superfoetation.

Other Evidence

A number of cases in which masses containing two foetuses of markedly different size and development were aborted are brought forward in support of the theory of superfoetation. In 1927 Riddel reported a case diagnosed as a missed abortion. At operation a three-and-a-half-

months macerated foetus and placenta were discovered, together with a healthy growing foetus of seven weeks. He suggests that in this case ovulation may have begun again after the death of the larger foetus. Willis (1929) cites a case in which two foetuses, estimated as being of nine and sixteen weeks' gestation respectively, were aborted. A single placental mass with two cords issuing from it excluded the possibility of a double uterus. Hansen (1932) reports a similar case, in which he estimates the gestation periods at one and three months; both foetuses were quite normal and appeared only recently dead. Other such cases are reported by Langmore (1863), Tyler Smith (1856), Gustetter (1918), Föderl (1932), Studdiford (1936), Radasch (1921), and others. In the case Radasch describes an aborted mass contained two foetuses of sixteen weeks' and forty days' estimated gestation in separate sacs. Neither showed any signs of maceration. He points out that the younger a foetus is the more rapidly does maceration take place, and he feels that his is a case of superfoetation, as a young foetus could not remain dead in the uterus for two months without showing marked maceration.

Superfoetation in Animals

There is much convincing evidence that superfoetation can take place in animals, though here also it must be regarded as an extremely rare event. Slonaker (1934) has produced perhaps the strongest evidence in its favour in his work with albino rats. These animals show periods of activity and alterations in the vaginal smear corresponding with ovulation. During pregnancy, which lasts twenty-two days, these characteristic signs of ovulation are absent, but in a few animals he observed periods of activity occurring between twelve and fifteen days after coitus. In these cases he presumed that ovulation was occurring during pregnancy, and accordingly left two such animals with the male till the twentieth day after the first coitus. Both animals gave birth to two normal litters, the first on the twenty-second, followed by a second litter on the thirty-seventh day after the first coitus. He feels sure that both were cases of true superfoetation. Numerous other examples of superfoetation in animals are reported. Buchanan Smith (1927) observed superfoetation in large black sows, in cases which he concludes cannot be explained by arrested development. He also cites cases occurring in sheep. Markee and Hinsey (1935) report a probable case in a cat. Other instances are cited by King (1913), who has also observed cases in albino rats.

Conclusion

We have described a case of which we think the most likely explanation is superfoetation. Unfortunately there was no scientific proof and no medical witness of the first birth, but from our observations we are both convinced that the case is a genuine one. A number of similar cases are described in the older literature but none in recent years. This we attribute to the fact that in modern times comparatively few cases escape the attentions of doctor or midwife.

An outline of some of the literature is given, showing evidence that ovulation may occur during pregnancy; that cases of abortions containing foetuses differing widely in size and development are seen from time to time; that cases are reported in animals.

Lastly, it should be remembered that since all possible circumstances are against superfoetation occurring, it is extremely rare, but there is an exception to every rule, and just because it has not occurred, or rather has not

been reported previous to a certain time, does not mean it can never occur, and it is only fair for those who would be dogmatic to consider such a case as this with an open mind.

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KERATOPLASTY

BY

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During the last few years much attention has been drawn to corneal grafting by numerous publications in ophthalmological and other journals. The experimental work of Tudor Thomas in England and Castroviejo in America has illuminated the subject considerably, but has perhaps tended on the whole to emphasize the difficulties of the procedure in the minds of medical men. The demand for the relief afforded by keratoplasty is relatively small in Western countries, and this, together with the comparative scarcity of good material for transplants, limits the field and tends to make the majority of ophthalmic surgeons in the West hesitate to adopt a measure which presents so many apparent difficulties.

Conditions are different in the Near and Far East, where the demand for relief by corneal grafting is considerable and suitable grafting material is easy to obtain. Under such circumstances it is only to be expected that simplification of technique and greater rapidity of execution should be aimed at and acquired, and although reports of cases and operative detail from Eastern Europe, India, and adjoining areas do not show up prominently in the literature, there is little doubt that in these areas much more practical experience has been obtained in performing corneal grafting on the human subject than elsewhere.

A Simple Technique

I am not familiar with the exact methods adopted by the various ophthalmic surgeons who freely employ keratoplasty in the East, but I have evolved a simple technique which appears to hold out as good a chance of success as the more elaborate methods described in recent literature. It has the advantage that it can be undertaken confidently by any ophthalmic surgeon of experience. I demonstrated this method in Melbourne at the Annual Meeting of the British Medical Association, but only on the rabbit, as a patient and donor were not available.

The trephine is used for both graft and bed. The nests of trephines made for me by Messrs. Down Brothers give a fair range in size. Although I have employed a 12-mm. trephine on two occasions my impression is that this is too big, not because the graft does not readily take, but because anterior synechiae are more likely to occur. The formation of an anterior synechia with the back of the graft junction is the complication most to be dreaded in keratoplasty. A 3-mm. trephine is too small except when one employs it for the graft method of treating fistula which I have described. The best average trephine diameter is perhaps 6 mm. There is no real need to have special nests of trephines, although the finish of the edge in specially made cylinders gives a bevel to the margin of graft and trephine hole. Cork borers, such as are found in a laboratory, are almost equally good. No other special instrument is required, although of course it is preferable to use fine needles, such as Barraquer's or Kalt's half-circle 7 mm. point to eye (maker's 12 mm.), and a delicate needle-holder such as that made for me by Weiss. The various steps in the procedure are briefly as follows.

The cornea chosen for grafting should preferably have a normal anterior chamber. (This is highly advisable in earlier attempts. Later the surgeon may consider what we refer to as reconditioning the anterior chamber before grafting, a much more difficult and tedious procedure.) The graft should for preference be obtained from a living human eye. I have no experience of grafts obtained post mortem. Eyes blind from glaucoma furnish quite good grafts, as do eyes removed in the course of exenteration for malignant disease. It appears to be unnecessary to choose a donor of the correct blood group. The donor and recipient are placed on adjoining tables and prepared in the usual way as regards surgical cleanliness. Akinesia and anaesthesia are established by novocain and adrenaline; pantocaine drops are given. The recipient's pupil should be small, the donor's wide, if this is possible. The necessary instruments for completing the enucleation or exenteration on the donor are left ready and an operator detailed for this work.

Preparation for Transplantation

Having decided on the disk diameter and the trephine to be used, the site is demarcated on the recipient's leucomatous cornea by a few rotations of the trephine. The stitches are then put in, taking the first bite at 6 o'clock just off the cornea, where an easy but firm grip can be taken with the point of suture, which does not produce conjunctival drag or puckering. One end of the thread is held; the needle with the other is carried across the cornea to 12 o'clock, where a similar bite is taken. The needle is then unloaded and the free end carried back to 6 o'clock and laid beside the end already in position. This manœuvre is repeated at points corresponding to 7.30 and 1.30, 9.30 and 4.30. It is well to do this clockwise and remember the order of placing the sutures. One observes how they will eventually be related to the graft by noting where they cross the demarcated area. The stitches are then pulled free of the demarcated area, the lids closed, and attention given to the donor, who now has a speculum *in situ* and is ready prepared.

Trephining the Donor

Here the same trephine is used in a corresponding position on the cornea. Good fixation of the globe is desirable. This can be effected simply by a T-shaped fixation forceps. The trephine must be used evenly and freely.