megaloblastic anaemia, with low serum B₁₂, unresponsive to parenteral cyanocobalamin therapy. Although the patient's serum vitamin B₁₂ level was raised from 20 to 1,000 μg./ml. by therapy, there was no clinical or häematological response. Further study of vitamin B₁₂ metabolism in this disease is clearly required.

Summary

A case of Di Guglielmo's syndrome has been described, in which megaloblastic anaemia was associated with low serum vitamin B₁₂ level. The case is compared with others previously described. The need for further study of vitamin B₁₂ metabolism is emphasized.

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Addendum

Since this paper was prepared work has been published (Boczarow, 1961) suggesting that penicillin will invalidate B₁₂ assay by the L. leichmannii method. Previous work by Dr. L. G. Bruce, in the clinical laboratories of the Victoria Infirmary, did not suggest this and estimations of serum vitamin B₁₂ levels in our patient were done by his methods, which differ slightly from those of Boczarow. The validity of the results was confirmed by further studies, which will shortly be published.

REFERENCES


Bruce, L. G. (1961): Personal communication.


Kay, A. W. (1953): Effect of Large Doses of Histamine on Gastric Secretion of HCl; An Augmented Histamine Test, Brit. med. J., ii, 77.


42 weeks in duration, and had terminated spontaneously. The delivery and puerperium in each instance had been normal. The birth weights were 8 lb. 16 oz. and 8 lb. 7 oz. respectively.

On examination, the patient looked somewhat pale. Pulse 104/min., regular. BP 116/60 mm. Hg. The uterus was at term, but was a little tense and slightly tender over a small area on the left and just below the umbilicus. The fetus was lying in the right occipito-lateral position and the presenting part was fixed in the brim. Fetal heart rate: 144/min.

Since admission to the hospital some 12 hours previously there had been a further loss of 12 oz. of blood p.v. Owing to unavoidable circumstances the patient had not been seen earlier, although blood and urine investigations had been performed. The hemoglobin estimation was 64% (9.5 g./100 ml.). A specimen of blood clotted within 3 minutes and the clot remained firm and stable for the next 24 hours. A catheter specimen of urine contained 80 mg. protein/100 ml.

A diagnosis of a minor degree of accidental ante-partum haemorrhage was made and a blood transfusion was started. A vaginal examination, performed in the operating theatre, revealed that the cervix was thick, soft and admitted two fingers. The vertex was presenting at the brim.

A wide sweep of the membranes off the lower segment and gentle stretching of the cervix were performed and then the forewaters were ruptured with a pair of Kocher’s forceps; 20 oz. of clear liquor were drained. Pethidin, 150 mg., had been administered to the patient just prior to the vaginal examination.

Subsequent Progress

Two hours later: Maternal pulse 92; fetal heart rate 156. Four hours later: Patient complaining of backache. Maternal pulse 90; fetal heart rate 154; BP 120/80. Five hours later: Patient getting contractions. Slight dark loss p.v. Twelve hours later: Contractions every 4 minutes. Maternal pulse 92; fetal heart rate 156; BP 160/90. Fourteen hours later: Fetal heart rate more than 180/min. Presenting part engaged. Urine: no albumin, no acetone. Vaginal examination showed the cervix to be four fingers dilated and vertex presented. Vertex presenting in the mid-cavity, in the direct occipito-posterior position. Small quantities of blood-stained liquor were draining.

It was decided to deliver by lower segment Caesarean section. A specimen of blood, taken an hour before operation, formed a firm stable clot within 4 minutes. A lower segment Caesarean section was performed under a general anaesthetic, consisting of nitrous oxide, ether, oxygen and scoline.

At laparotomy a small quantity of dark red free blood was found in the peritoneal cavity; both broad ligaments and the entire upper segment were a deep plum colour, in distinct contrast to the lower segment, which appeared normal in hue.

A severely asphyxiated female infant, weighing 8 lb. 10 oz. was delivered. Ten minutes after delivery, after the administration of intra-gastric oxygen, regular respirations were established.

Approximately 20% of the lower edge of the placenta had separated and there was a retro-placental clot measuring 20 oz. The placenta, otherwise, appeared quite normal. The patient had received 3 pints of blood altogether.

For the first seven days the infant exhibited hypotonicity and had attacks of intermittent cyanosis and was unable to take feeds. A subdural tap revealed no haemorrhage and a lumbar puncture showed slightly xanthochromic fluid under normal pressure. Laboratory investigation showed the cerebral spinal fluid to be normal.

On discharge on July 31, 1961, the infant had made a complete recovery and weighed 8 lb. 8 oz.

On August 11, 1961, the infant was seen by the paediatrician, who stated that the baby had made a good recovery and was behaving normally in every way. The infant weighed 11 lb. 14 oz.

The patient’s haemoglobin on the fourth post-operative day was 60%. She was transfused with a further pint of blood. Her recovery was normal. On discharge the haemoglobin was 80%.

Case 2

A multigravida, aged 21. In 1960 she gave birth normally to a male child, weighing 7 lb. 4 oz., at the City of London Maternity Hospital, where she was booked for this second pregnancy. The haemoglobin remained low (67% to 70%), otherwise she was well and last seen at the 38th week.

One day before term, on October 30, 1961, she awoke at 6 a.m. with normal labour pains. Four and a half hours later she was admitted and found to be bleeding. The pains had changed and were becoming constant; BP 80 mm. Hg, pulse 95. The uterus was woody hard. The fetal heart rate was 120. The urine contained 200 mg. albumin/100 ml. The blood contained negligible amounts of fibrinogen, under 50 mg./100 ml. Triple strength plasma was given.

One and a half hours after admission 2 pints of triple strength plasma had been given. The blood pressure was 104 mm. Hg, pulse 130, fetal heart rate 180. Laparotomy was performed. The uterus was tense and plum coloured; there were many thromboses. Blood-stained liquor flowed out through the incision together with a few clots. The head was jammed down into the pelvis and difficult to deliver.

The baby was a girl who cried at birth and weighed 6 lb. 10 oz. A third of the placenta was separated. There were retroplacental clots in the fundus. There was an estimated amount of 3 to 4 pints of blood in the uterus. The blood was not clotting well; 287 mg. of fibrinogen had been given. A few hours after the operation the blood was clotting well. The blood fibrinogen was 120 mg./100 ml. Five pints of blood had been given.

Progress of mother and baby was satisfactory.

Discussion

Two cases of abruptio placenta associated with a Couvelaire uterus are reported. Few, if any, such cases have been reported where a living infant was delivered. The Couvelaire uterus is regarded as a manifestation of the blood-clotting defect associated with abruptio placenta. In the first case it is postulated that had delivery been delayed the full-blown picture of afibrinogenaemia would have resulted. It is of interest that the clot-observation test revealed a firm stable clot, despite the appearances at Caesarean section. It is regretted that the fibrinogen estimations had not been done.

The alarming uterine conditions sometimes seen and studied at laparotomy merit thought. There seem to be two factors. One is mechanical: in the really severe cases the baby—usually the head—acts as a plug to the cervix and all the blood is pent up in the fundus. Rupture of the membranes fails to release the extra-chorionic blood. If the bleeding continues, the uterus becomes increasingly distended and is unable to contract. If the uterus is less distended or disorganized, then the cervix dilates...
and uterine contractions occur. In the severe cases it is erroneous to say that the uterus is atonic. Hysterectomy is rarely required and the most strikingly thrombosed and blood-infiltrated uterus will contract and behave normally, helped by prompt intravenous ergometrine. That experience was obtained before the ample supply of fibrinogen.

The other factor, not fully understood, is biochemical and hematological: as always, blood lost in any quantity from the circulation must be replaced as soon as possible. This is well known and massive replacement, 6 to 15 pt., not uncommon. The need for blood nearly always exceeds the estimated requirement.

The severe cases are characterized by continued intra-uterine bleeding. A larger volume of blood is lost from the circulation than the observer realizes. Finally and suddenly the patient collapses.

From these severe cases there are all gradations back to those cases which have a slight inconsequential bleed, which may be almost overlooked. How soon is blood in the uterus changed into dark clot? Are all the accompanying changes known? How much intra-uterine blood can be completely absorbed without leaving a trace? How often does intra-uterine bleeding occur?

The condition, even when severe, may well be more common throughout the country than is generally supposed. That it threatens the life of the mother—as placenta praevia used to—maternal statistics show.

The cause remains an open question.

Albuminuria and raised blood pressure—relative to the degree of clinical shock—are usually present. There is no evidence that they are causal, otherwise we would expect concealed ante-partum hemorrhage to be far more common and dreaded as an expected complication of severe pre-eclamptic toxæmia. When we admit a severe case of pre-eclamptic toxæmia we fear fits rather than ante-partum hemorrhage.

It would seem reasonable to conclude that there is some unknown, possibly chemical or anatomical, factor or factors which can cause raised blood pressure, albuminuria and/or retro-placental bleeding.

For nigh on two centuries the division between accidental and unavoidable ante-partum hemorrhage has been stressed in teaching and practice. It could be that the division has become, over the years, too rigid in minds and books. What was a pioneering observation could become a brake on progressive thought. Both unavoidable and accidental uterine hemorrhage were maternal killers. So far as placenta praevia is concerned, progress surged forwards when the vaginal approach for both diagnosis and treatment gave way to early expert abdominal approach. There followed improvement for mother and baby.

Accidental ante-partum hemorrhage, according to some current teaching, must always be treated per vaginam; dogmatic assertions, such as ‘Caesarean section has no place in the treatment of accidental ante-partum hemorrhage’, are sometimes bandied about by closed minds.

There may be a uterine world of difference between retroplacental hemorrhage, when the placenta is located symmetrically at the fundus, compared with its location symmetrically over the cervix. But those are not the commonest locations or syndromes.

Most cases commence at home, out of the blue, usually in a patient who has been considered antenatally normal and safe. In the early case there may be few clinical signs.

Course

The course cannot be forecast at the onset. The time factor between onset and danger to baby or mother is vitally important.

Intra-uterine hemorrhage can be one of the gravest emergencies in obstetrics, threatening the life of the baby in two to three hours and that of the mother in five to six hours. The clinical features vary so widely that statistics can mislead.

The whole problem should be approached afresh, under the heading of intra-uterine hemorrhage throughout pregnancy.

New methods and techniques for studying intra-uterine conditions are urgently required.

REFERENCES


Life from a Couvelaire Uterus

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