REFERENCES

- Blair, W., Brown, W. Kerr, Caldwell, W. F., Clarke, A., Dunn, S., Primrose, D., Stewart, T. F., Grist, N. R., and Sommerville, R. G. (1959). *Brit. med. J.*, 1, 422.
- Chanock, R. M. (1956). *J. exp. Med.*, 104, 555.
- Parrott, R. H., Cook, K., Andrews, B. E., Bell, J. A., Reichelderfer, T., Kapikian, A. Z., Mastrotta, F. M., and Huebner, R. J. (1958). *New Engl. J. Med.*, 258, 207.
- DeMeio, J. L., and Walker, D. L. (1957). *J. Immunol.*, 78, 465.
- Dubowitz, V. (1958). *Lancet*, 1, 140.
- Dunbar, J. M., Jamieson, W. M., Langlands, J. H. M., and Smith, G. H. (1958). *Brit. med. J.*, 1, 913.
- Gardner, P. S. (1957). *Ibid.*, 1, 1143.
- Grist, N. R. (1958a). *Hlth Bull. (Edinb.)*, 16, 5.
- (1958b). *Ibid.*, 16, 56.
- Sommerville, R. G., and Carson, H. G. (1957). *Brit. med. J.*, 1, 378.
- Horne, W. A. (1957). *Influenza 1957: Report on the Epidemic in Glasgow*. Corporation of Glasgow, Health and Welfare Department.
- Kuroya, N., Ishida, M., and Shiratori, T. (1953). *Yokohama med. Bull.*, 4, 217.
- McGill, R. J., and Goodbody, R. A. (1958). *Lancet*, 1, 320.
- Morgan, D. M., and Pickup, J. D. (1958). *Brit. med. J.*, 1, 263.
- Stephens, J. H., and Cappell, D. F. (1958). *Lancet*, 1, 381.

ANAEMIA AND POLYCYTHAEMIA IN UNIOVULAR TWINS

BY

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The simultaneous occurrence of anaemia and polycythaemia in newborn monozygotic twins was first reported by Herlitz (1941-2), who described two such instances. Klingberg *et al.* (1955) and Bergstedt (1957) have reported similar cases, but none has so far been reported in Great Britain. In the past three years the syndrome has been recognized on four occasions in the Glasgow Royal Maternity and Women's Hospital. It seems worthy of comment, since in severe cases prompt treatment may be required. The findings in the cases previously reported are summarized in Table I, and in

TABLE I.—Summary of Previously Reported Cases

Author	Weight in g.	Sex	Hb	R.B.C. mil./c.mm.	Outcome
Herlitz (1941-2)	1,580	M	50%	1.94	Well at 6 years
	2,580	M	160%	7.02	
	3,030	F	200%	6.86	Died at 2 months. "Bronchitis"
	2,220	F	65%	2.27	
Klingberg <i>et al.</i> (1955)	2,690	F	25.2 g./100 ml.	7.47	Uneventful
	1,770	F	3.7 "	1.85	
Bergstedt (1957)	3,030	F	23.6 "	5.90	Well at 5 months
	1,810	F	13.6 "	3.90	

Table II the peripheral blood values in the present series are given. The following is a short history of the births and clinical findings in the four sets of twins.

Case 1

This patient, a 3-gravida, was delivered spontaneously of twin boys after 37 weeks' gestation. She was known to be rhesus-negative, but no antibodies had been detected. Apart from hydramnios, the pregnancy had been uneventful and the onset of labour was spontaneous.

The first twin, an assisted breech delivery, cried lustily at birth and weighed 2,570 g. The second child, a spontaneous vertex delivery, cried shortly after birth and weighed 2,680 g. This infant looked pale, the spleen tip was palpable, and the liver was enlarged to three fingerbreadths below the costal margin. Examination of the peripheral blood revealed a severe anaemia with a reticulocytosis, and many erythroblasts were to be seen in the film. Since the

mother was known to be rhesus-negative, erythroblastosis foetalis was the presumptive diagnosis. The Coombs test on the cord blood was, however, persistently negative. Despite this finding, an exchange transfusion of 500 ml. was carried out.

The results of the examination of the cord blood of the first twin showed that he also had a negative Coombs test but that the haemoglobin level was remarkably high. Full blood examination confirmed the polycythaemia, but there was no excess of reticulocytes and the blood film was normal. Venesection was considered, but thought to be unnecessary.

The single placenta, which had been retained *in utero* and was therefore not available for inspection when the anaemia was discovered, was quite striking in appearance, in that one half was grossly congested with tortuous vessels, while the other half was pale with collapsed vessels.

Both babies made good progress. The plethoric infant had congested hands and feet for several days, and even on the eleventh day of life the haemoglobin remained very high at 20.5 g. per 100 ml.

Case 2

This patient, a nullipara, was well throughout her pregnancy, went into labour spontaneously after 34 weeks' gestation, and had normal vertex deliveries of twin girls. The first infant weighed 1,470 g. and on admission to the

TABLE II.—Peripheral Blood Values in the Present Series

Case No.	Weight in g.	Sex	Peripheral Blood			
			Hb (g./100 ml.)	R.B.C. (mil./c.mm.)	Retic. (%)	Film
1 {	2,570	M	21.8	8.66	2.3	Occasional erythroblasts Very numerous erythroblasts
	2,680	M	7.7	2.20	19.0	
2 {	1,470	F	20.0	5.57	2.2	Occasional erythroblasts Numerous erythroblasts
	1,080	F	8.3	2.28	15.0	
3 {	3,230	F	19.2	4.61	3.5	No abnormality " "
	2,190	F	14.1	4.42	7.3	
4 {	2,410	F	14.7	4.25	—	— —
	2,490	F	21.8	7.00	—	

premature baby unit was intensely cyanosed and had generalized oedema. The spleen was not palpable, but the liver was enlarged to three fingerbreadths below the costal margin. It was noted that the infant was plethoric compared with its twin, and this was confirmed by examination of the peripheral blood (Table II). Although the baby's colour improved with oxygen, respirations were never well established. At 3 hours her condition deteriorated. Venesection of 30 ml. of blood did not produce any improvement, and she died four hours after delivery. At necropsy there was congestion of all organs, the brain showing the presence of numerous small haemorrhages in the subependymal region.

The second twin weighed only 1,080 g. and cried shortly after birth. She was strikingly pale compared with her sister, and examination of the peripheral blood confirmed the anaemia. The Coombs test was negative.

Four and a half hours after delivery a transfusion of 40 ml. of blood was given into the umbilical vein. Her condition was satisfactory, considering the weight, until she was 13 hours old. Thereafter she had frequent apnoeic attacks and died at 18 hours. At necropsy there was some congestion of cerebral vessels, haemorrhage in the left adrenal gland, and widespread hyaline membrane formation in the lungs.

The placenta in this case was similar to that of the first, one side being grossly congested while the vessels of the other half were collapsed.

Case 3

This patient, a 4-gravida, was well throughout her pregnancy and went into labour spontaneously at term. The first twin was a spontaneous vertex delivery and cried at birth. She weighed 3,230 g. and was admitted to the well-baby nursery. At the age of 12 hours she was transferred

to the paediatric unit because she had become chilled and had developed generalized cyanosis. She improved quickly with the administration of oxygen, but the extremities remained cyanosed. Next morning it was appreciated that she was plethoric compared with her twin, and this was confirmed by examination of the peripheral blood (Table II).

The second twin weighed 2,190 g. at birth. She was an assisted breech delivery and was lethargic, but she improved quickly on admission to the premature-baby unit. On the second day examination of the peripheral blood revealed a mild degree of anaemia, for which no treatment was required. In neither twin were the departures from normal as severe as in the previous cases, and further progress was satisfactory. Unfortunately, by the time the condition was recognized the placenta was not available for inspection.

Case 4

This patient, a 6-gravida, was admitted to hospital at 35 weeks' gestation with a mild pre-eclamptic toxæmia. She went into labour spontaneously at 36 weeks and had two normal vertex deliveries.

The first twin weighed 2,410 g. and cried at birth. On the second day examination of the peripheral blood showed a mild anaemia, but no treatment was required.

The second twin weighed 2,490 g. at birth. At the age of 24 hours it was noted that her hands and feet were cyanosed, and examination of the blood showed a considerable polycythaemia.

Both infants were discharged home well. Regrettably, the placenta from this case had also been destroyed.

Discussion

In Case 1 the clinical picture was confused by the fact that the mother was known to be rhesus-negative. The infant was obviously in urgent need of blood, and, in spite of the negative Coombs test, the likeliest diagnosis at that early stage seemed to be haemolytic disease of the newborn: hence an exchange transfusion was done. In retrospect, a simple blood transfusion would have been just as effective as the exchange, but it is worth while noting that in the case described by Klingberg *et al.* (1955) an exchange transfusion was also given. Chown (1954) also carried out an exchange transfusion on an infant in whom the severe degree of anaemia at birth was later proved to have been due to bleeding of the foetus into the mother's circulation.

Observation of the first Glasgow case made it possible for the second to be readily recognized, and simple transfusion seemed the appropriate treatment. Venesection did not improve the plethoric twin, but both these infants were handicapped by their low birth weight. In the other two cases seen since no treatment was required.

It can be assumed that in all the cases described there must have been some direct communication between the circulations of the twins. Bergstedt (1957) confirmed this in his case by the injection of milk into one of the cords, with the result that both halves of the placenta became filled with milk. Klingberg and his co-workers also demonstrated arteriovenous shunts between the two placental circulations by perfusion of an artery in each cord with contrasting coloured fluids. The placenta in their case showed one half to be moderately red, without engorged vessels, and this they suggested supplied blood to the polycythaemic twin. The other half of the placenta was smaller and purple-red in colour with markedly distended veins; this half supplied blood to the anaemic twin.

That arteriovenous shunts occur in the placenta in uniovular twin pregnancies has been known for many

years. Potter (1952) states that it is "common"; Browne and McClure Browne (1955) maintain that there is always a communication, and refer to the work of Schatz (1884) on the subject; Schatz believed that the anastomoses may be superficial or deep, and that, while the superficial ones may be either arterial or venous, the deep ones are always arteriovenous. These anastomoses are of significance because their presence in uniovular twins may explain why the death of one foetus is more common in uniovular than in binovular twins.

However, as anaemic and polycythaemic twins occur but rarely, some other factor must be present to produce the condition. One must postulate some obstruction to the return of the blood to the placenta from the plethoric infant, or obstruction of entry of blood in the anaemic one. This would agree with the placental findings—the engorged placenta of the anaemic twin and the absence of engorgement in the placenta of the polycythaemic one.

The nature of this obstruction is not easy to determine. In six of the eight cases summarized in Tables I and II the polycythaemic twin was the first to be born. Blood loss at the time of delivery is not recorded and is unlikely to be a factor. Pressure on one or other cord, or on a superficial vessel in the placenta, would produce such a condition, but proof is lacking, and, in any event, would be difficult to find. It would seem that the condition probably exists for some hours at least *in utero*, because of the presence of the reticulocytosis in the anaemic twin (Table II). More detailed study of the placentas might provide further information.

The syndrome, especially in a mild form, is probably more common than is realized, and the recognition of the severe case is important because treatment by transfusion may be urgently required. Neligan and Russell (1954), discussing foetal anaemia due to blood loss after incision of the placenta during caesarean section, maintain that the exsanguinated baby may appear deceptively well until shortly before death. They suggest that a transfusion is required if the peripheral blood haemoglobin level falls below 13.3 g. per 100 ml. in the first 24 hours of life. Presumably this level may be taken as a guide in all cases of acute anaemia in the newborn, whatever the cause.

Summary

Four cases of monozygotic twin pregnancy are described, in each of which one twin exhibited a polycythaemia and the other an anaemia.

In two of the four cases the anaemia was severe enough to warrant blood transfusion.

The aetiology of the syndrome remains obscure, but must depend on direct communication of the two foetal circulations.

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REFERENCES

- Bergstedt, J. (1957). *Acta paediat.*, **46**, 201.
- Browne, F. J., and McClure Browne, J. C. (1955). *Antenatal and Postnatal Care*, 8th ed. Churchill, London.
- Chown, B. (1954). *Lancet*, **1**, 1213.
- Herlitz, G. (1941-2). *Acta paediat.*, **29**, 211.
- Klingberg, W. G., Jones, B., Allen, W. M., and Dempsey, E. (1955). *Amer. J. Dis. Child.*, **90**, 519.
- Neligan, G. A., and Russell, J. K. (1954). *J. Obstet. Gynaec. Brit. Emp.*, **61**, 206.
- Potter, E. L. (1952). *Pathology of the Fetus and the Newborn*. Year Book Publishers, Chicago.
- Schatz, F. (1884). *Arch. Gynäk.*, **24**, 337.