It was obviously important to find out whether the patient had at any time between the delivery and the follow-up investigation been pregnant. This seemed impossible. She had attended a welfare centre regularly. Husband and wife confirmed the use of birth control (by male sheath) because they did not desire a second child until they no longer had to live in the same house as a relative suffering from pulmonary tuberculosis.

The patient gave a history of regular menses and had received no injections of any kind either ante- or post-natally, and had never received a transfusion.

Case 2

A woman aged 22 was delivered of a living healthy, full-term baby on September 19, 1948. The infant showed no signs of haemolytic disease of the newborn. The infant's blood type was Group A rhesus-positive.

The maternal serology antenatally was Group A rhesus-negative, the serum containing no anomalous antibodies. A specimen of blood obtained on the eighth day after delivery gave identical results. A further specimen, however, obtained on June 22, 1949, gave definite evidence of the presence of a potent anti-rhesus antibody, again acting more strongly on albumin-suspended rhesus-positive cells. The titre was moderately high (1:8). The serological behaviour of the antibody was typical, no reaction occurring unless the test cells contained the D antigen.

This patient had shown no sign of a further pregnancy, her menses having been regular since the cessation of lactation three months after delivery. She had received no injections ante- or post-natally or transfusions at any time of her life. Her husband was Group O rhesus-positive and most likely genotype CDe/cde.

Discussion

The serological findings in these two cases are clear-cut and indicative of rhesus immunization. The fact that the antibody reacted better with albumin- than with saline-suspended cells might perhaps be due to its comparatively low titre. We share with several other workers the experience that sometimes an "albumin" antibody can change into a saline agglutinating antibody just before delivery. The contention, therefore, that a blocking antibody is always a sign of hyperimmunization might perhaps be open to discussion.

It is difficult to understand why antibodies in these two patients developed so late after delivery. In both, the antigenic stimulus must have been the first pregnancy, and, so far as can be established, there was no further antigenic stimulus. That antibodies can develop to a stimulus a long time after its first application is a not uncommon experience in immunology—for example, undulant fever. Further investigations are in progress to elucidate this problem in greater detail. As a practical application of our findings, if they can be confirmed on a larger series, it might perhaps be advisable to investigate for the presence of anomalous antibodies the sera of all primiparae contemplating a second pregnancy.

Summary

In a follow-up of 62 rhesus-negative primiparae with rhesus-positive babies, two were found who, after intervals of two years and of nine months respectively, had developed anti-rhesus antibodies in their serum without a further intervening pregnancy or other antigenic stimulus.

We are greatly indebted to the technical staff of the Blood Transfusion Service in Birmingham and to Sister Lyons, of Sorrento Maternity Hospital, for the valuable help they have given in testing the many specimens and in following up the patients.

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MONGOLISM IN ONE OF TWINS

BY

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Graham (1950), reporting a case of mongolism in one of twins, drew attention to the comparative rarity of such cases in the British literature. The largest series so far published seems to be that of Warner (1949), who collected 101 cases from the literature and recorded one further case in which an older sib was also a mongol. Of these 102 sets of twins, one child was affected in 82 cases and both children in 20.

It is well recognized that there is a considerably increased incidence of congenital cardiac defect among mongols. Wylie (1949) quotes 22 examples in a series of 138 mongols (16%); Benda (1947) states that up to 75% of mongols dying in the first two years of life have a congenital cardiac defect, as have 35% of those surviving. Post-mortem records of the Hospital for Sick Children, Great Ormond Street, yielded evidence of congenital cardiac abnormality in 28 of 63 mongols (Evans, 1950). While various malformations are encountered, persistent ostium atrio-ventricular commune is especially apt to be associated with mongolism (Abbott, 1936). Robinson (1941), in a series of 39 cases of this defect collected from the literature, states that about one-half of these were in mongols. Taussig (1947) emphasizes the likelihood that a mongol presenting signs superficially resembling ventricular septal defect, and without cyanosis, is suffering from persistent atrioventricularis communis. There were, however, only four examples of this in Evans's series, the commonest anomaly being in fact ventricular septal defect.

Case Report

Helen B. was born on November 20, 1949, at full term, the first of twin girls. The mother was aged 35; her health was good during pregnancy, apart from a cystocele which was troublesome during the first trimester. The father's age was 36. The family and personal histories of both parents were, so far as is known, good. There are two older (normal) daughters, now aged 4 and 7 years. Helen was delivered as a vertex, and the second (normal) twin as a breech; each weighed approximately 5½ lb. (2.5 kg.) at birth. A suggestion of mongoloid physiognomy was noted in Helen at birth, but was discounted to some extent by the medical attendant because

of a strong family resemblance between the infant, the father, and the two elder children.

From birth breast-feeding was difficult, and soon became impossible; she was given expressed breast milk for four weeks, and afterwards a mixture of cows' milk and water was substituted. She never thrived so well as her twin, and on first examination weighed only 9 lb. (4.1 kg.), as against 12 lb. (5.4 kg.). The mother noticed that she was less lively than her twin, was easily made breathless, and sometimes "went blue." She vomited slightly from birth, but at the time of first attendance this was not causing any anxiety.

The child was examined in the out-patient department of the Royal Belfast Hospital for Sick Children on January 20, 1950, when the diagnosis of mongolism was confirmed and its significance explained to the mother. The accompanying photograph was then taken.

She was again brought to the out-patient department on January 28 with a history of severe vomiting of a projectile type. On examination the pylorus was easily palpable and thought to be thickened. In view of the presence of a systolic murmur to the left of the sternum a chest radiograph was taken, which showed considerable generalized enlargement of the heart. A dietetic regimen and "pylostropin" tablets were used until admission could be arranged, but at the time of admission (January 31) she was vomiting as copiously as ever. Surgical treatment of the pylorospasm was decided upon, despite the known multiple hazards present.

Mr. J. A. W. Bingham opened the abdomen by a right transverse rectus-cutting incision under "soneryl"-atropine-vinyl-ether anaesthesia on February 2. Bleeding was slight and the blood dark. Before the peritoneum could be incised the heart stopped suddenly. Cardiac massage, artificial respiration, and oxygen inhalation produced only transient response.

Post-mortem Examination

At the post-mortem examination, carried out by Dr. R. J. Marshall, a poorly nourished female infant with the typical facies of mongolism was seen. The heart was considerably enlarged: normal epicardium; ductus arteriosus closed; the great vessels entered and left the heart in normal arrangement. The right auricle was much enlarged, and the wall markedly trabeculated. The interauricular septum was thin, terminating in a crescentic free edge just proximal to the auriculoventricular ring. The septum was fenestrated, and the foramen ovale closed. Anterior and posterior valve cusps were present, but were abnormal in that they continued uninterruptedly across the septal defect, the distal halves serving as mitral cusps.

The right ventricle was also markedly enlarged, and there was gross defect of the interventricular septum, which was rudimentary and present only at the apex of the organ. The ostium between the edges of the auricular and ventricular septa measured 2 by 1.5 cm.; thus there was virtually a common ventricle, the right half of which was larger than the left.

The stomach and duodenum were essentially normal in size and shape, and, although the pylorus was somewhat thickened, no congenital hypertrophy was noted. The lungs showed patchy interstitial pneumonia with mononuclear infiltration of the alveolar walls and comparatively little alveolar exudate. The liver and spleen showed the persistence of haemopoietic tissue.

Summary

A case of mongolism with persistent ostium atrioventriculare commune in one of twins is recorded. Pylorospasm with intractable vomiting, inanition, and death under anaesthesia exemplify the precarious hold on life which is the inheritance of most mongols. The nature of the cardiac anomaly was such as to preclude lengthy survival in any event.

We are indebted to Dr. P. P. Jackson for the obstetrical details, and to Mr. D. Mehaffy, A.R.P.S., for the photograph.

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Medical Memoranda

A Vestigial Tail

Many cases of children born with a tail have been reported in the last seventy years, but very few in the English literature. The following is the description of one.

CASE REPORT

The patient first attended hospital on May 11, 1949, at the age of 11 months. He had been born with a tail-like appendage, but his mother had not sought advice before, as she thought that nothing could be done until the child had dispensed with napkins. However, the increase in size of the tail at last made her come to hospital. There was no family history of any like anomaly. The child was a firstborn, the pregnancy and delivery had been without incident, and the child had developed normally since birth. There were no bowel or urinary symptoms. On examination he was found to be a healthy normal child, apart from his caudal appendage. The tail was attached to the right posterior anal margin (see illustration) and could be freely moved by the child. It was extensible and when fully pulled out measured just over 3 in. (7.5 cm.). When released it would contract, curl on itself, resembling the tail of a pig, and then relax and flick from side to side.

Photograph showing the vestigial tail at 11 months.

No bone or cartilage could be palpated in the tail. It was covered with normal skin. The anus freely admitted the examining finger, and the sphincter was of normal tone. X-ray examination of the pelvis showed no bony abnormality.

On May 26 the tail was excised by an incision encircling the base. The wound was soundly healed eight days later.

The specimen was reported on as follows: "A finger-like process 7 cm. in length and 1.5 cm. in diameter at the base. It was covered with hair-bearing skin and its core consisted mainly of fibro-fatty tissue with a small eccentric nodule of 'cartilaginous' consistency. *Microscopy*:—The external lining was of epidermis with normal skin appendages. Most of the core was formed of striated muscle bundles. Also present were fat, fibrous tissue, blood vessels, and nerves."

COMMENT

Bartels (1884) traced 116 recorded observations on tails in man. He found references as far back as the writings