Short reports

medroxyprogesterone acetate treatment have been monitored by plasma testosterone, FSH, and LH estimations. The results suggested a partial suppression of the tumour's influence, the mechanism of which is discussed.

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REFERENCES


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Bone changes in congenital cytomegalic inclusion disease

We report the case of a female infant born with cytomegalic inclusion disease, in whom bone changes were present in x-rays of the tibiae and femora. She was the mother's first child and was born 8 days postmature after a normal delivery; birthweight 2.52 kg. Many infarcts were seen in the placenta. The mother had had a feverish illness at the 7th month of gestation, diagnosed by the family doctor as influenza. No drugs had been given.

Case report

The infant became jaundiced on the second day, serum bilirubin was 6-1 mg/100 ml, Hb 25 g/dl and the Coombs's test was negative. The liver was palpable 2 cm below the costal margin and the tip of the spleen was palpable. During the following days the jaundice became deeper and greenish in colour and the stools became pale. She was transferred to this hospital on the 9th day.

On admission the jaundice was marked, the liver palpable 3 cm below the costal margin, and the tip of the spleen palpable. Total serum bilirubin was 13.5 (unconjugated 6.8) mg/100 ml, serum alkaline phosphatase 10 units, thymol flocculation positive, thymol turbidity 4.0 units, zinc turbidity 4.5 units, SGOT 110 K units/ml, SGPT 73 K units/ml. The urine contained no pus cells or reducing substances, but bile, urobilin, and urobilinogen were present. Repeated specimens were examined for cytomegalic inclusion bodies with a negative result. The blood culture and Wassermann reaction were both negative. Serum immunoglobulins—IgG 1109 mg/100 ml (normal), IgA 10 mg/100 ml, and IgM 32 mg/100 ml. X-rays of chest and skull normal. X-rays of the legs showed oval translucencies at the distal ends of both tibiae and faint longitudinal striations at the distal ends of both femora (see Fig.). At the age of 2 months the appearance had returned to normal.

Blood was collected from mother and infant for viral studies and to exclude toxoplasmosis. At 12 days cytomegalytic titre was 1/512 (mother and infant); at 11 weeks 1/128 (infant); at 7 months 1/64 (infant).

Many specimens of urine were cultured for viral infections and were negative until the infant was 8 months old, when cytomegalovirus was cultured.

Progress. The jaundice gradually disappeared and by the age of 3 months the total serum bilirubin was less than 1 mg/100 ml. The liver, however, was still enlarged and firm and could be felt 3 cm below the costal margin; the spleen was still palpable. A liver biopsy showed that 'a giant cell hepatitis was present with minimal fibrosis' (Dr. J. M. Bouton).

Steady improvement followed and by the age of 3 years the liver and spleen were normal in size and the liver function tests were also normal. Examination of the central nervous system showed minimal cerebral dysfunction with slight clumsiness of movement of the limbs. Intellectual assessment at the age of 5 years was satisfactory, the IQ being 102 on the Stanford-Binet Scale.

Discussion

There have been numerous reports of bone changes in newborn infants with congenital rubella syndrome. The principal changes described are alternative dense and radiolucent striations in the metaphysis of long bones, particularly at the distal ends of the femora (Singleton et al., 1966). More recently similar changes were found in a patient with cytomegalic inclusion disease (Graham, Thal,
FIG.—Rarefaction at distal ends of both tibiae: longitudinal striations at the distal ends of the femora present in the x-ray film are not visible in the photograph.

and Wassum, 1970). The appearance of the bones improved and was normal at 5 weeks. It was indicated that such skeletal changes are due to interference with normal endochondral bone formation, not to osteomyelitis. Reporting a further case, Merton and Gooding (1970) suggested that similar bone changes might be expected to occur in association with other congenital viral infections. There is little doubt that the abnormal radiological appearance of the bones in the case presented here was due to cytomegalic inclusion disease.

**Summary**

A case of congenital cytomegalic inclusion disease with x-ray changes in bones is described. The x-ray changes were considered to be due to disturbance of endochondral bone formation and not due to viral osteomyelitis.
Coexistent coeliac disease, diabetes mellitus, and hyperthyroidism

About one in a thousand schoolchildren in Great Britain have diabetes mellitus. Coeliac disease and thyrotoxicosis are less common. The child described below had all three, the first time that such a combination has been reported in childhood.

Case report

A girl was born in July 1959. Until the age of 6 years she was well. At this time she began to get frequent bouts of abdominal pain and loose stools. Investigations showed an iron deficiency anaemia. This was treated with oral iron but the response was poor.

When 12 she complained of irritability, weight loss, polyuria, and polydipsia. Diabetes was diagnosed and confirmed biochemically. At this time, her refractory anaemia was investigated with the following results: Hb 11 g/dl; serum iron 51 μg/100 ml; serum folate 0·8 ng/ml; faecal fat excretion 8·25 g/24 h; blood xylose levels after 9·69 g xylose orally: 30 min 8·5 μg/100 ml, 1 hour 17 μg/100 ml; reticulin and thyroid antibody titres, negative; bone age 8 years 6 months (chronological age 12 years). The jejunal biopsy appearance (Fig. 1a) was that of subtotal villous atrophy. Intestinal malabsorption due to coeliac disease seemed to be the most likely cause of her long-standing symptoms.

For one year subsequently she thrived on a gluten-free, carbohydrate controlled diet with vitamin supplements. The diabetes was controlled with daily injections of lente insulin. Her weight gain velocity increased as shown in Fig. 2.

Early in 1973 she noticed that her stool frequency was increasing, and that she was experiencing abdominal pain unlike that which occurred when she ate gluten-containing foods. The rate of her weight gain diminished, and on direct questioning she admitted to heat intolerance, lassitude, and a voracious appetite. Physical examination showed a diffuse goitre over which a bruit could be heard. She had a tachycardia that persisted during sleep, moist palms, and a fine tremor.

There was neither exophthalmos nor lid lag, nor signs of diabetic retinopathy or neuropathy. Investigations confirmed the clinical diagnosis of thyrotoxicosis: serum T₄ 15·9 μg/100 ml, PBI 13·8 μg/100 ml, T₃ uptake 60%. Thyroid antibodies: thyroglobulin 1:100; microsomes, strongly positive IgG.

At this point it was discovered that her paternal uncle and grandfather both had thyrotoxicosis. Features of the uncle's condition included a high titre of circulating thyroid antibodies and poor control using drug therapy. He was eventually operated on. The grandfather responded well to drug treatment.

The present child was treated with carbimazole. The first sign of response was a decline in the insulin requirements for diabetic control. At this time, her intestinal function was reinvestigated with the following results: Hb 13·4 g/dl; blood xylose levels after 12·4 g xylose orally: 30 min 37 μg/100 ml, 1 hour 47 μg/100 ml; faecal fat excretion 7·05 g/24 h; sweat test normal; jejunal amylase and lipase activity normal; stool trypsin activity
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