Short reports

Value of 1-hour blood-xylose test in diagnosis of childhood coeliac disease

Rolles and his co-workers (1973, 1975a,b, 1976) have emphasised the value of the 1-hour blood-xylose test as a diagnostic tool in childhood coeliac disease, while others have questioned its reliability (Sladen and Kumar, 1973; Lamabadusuriya *et al.*, 1975). Our aim was to evaluate the test in a large group of control patients and in children with untreated coeliac disease.

Patients and methods

There were 46 children with untreated coeliac disease. None had received a gluten-free diet before the investigation. Jejunal biopsy was performed in all, and showed subtotal atrophy of the mucosa. All made an unequivocal clinical response to a gluten-free diet. The average age of the patients was 2.5 years, range 5 months to 12.7 years. 72% were under the age of 1.5 years.

The control group was made up of 102 patients with various gastrointestinal disorders (Table). Jejunal biopsy in all showed normal morphology. The average age of this group was 5.7 years, range 4 months to 15.8 years.

Table Diagnosis in 102 control patients

Suspected coeliac disease, unconfirmed	46
Failure to thrive due to extraintestinal reasons	28
Chronic nonspecific diarrhoea	13
Anorexia nervosa	4
Hiatus hernia	2
Isolated lactase deficiency	3
Selective IgA deficiency	1
Intestinal lymphangiectasia	1
Crohn's disease	1
Ulcerative colitis	1
Chronic constipation	1
Salmonella infection	1

The blood-xylose test (Rolles *et al.*, 1973) consisted of 5 g D-xylose in 100 ml water given to the fasting patient; exactly 1 hour later the venous blood sample was drawn. D-xylose was measured by the method of Roe and Rice (1948) as modified by Colombo (in Schaad *et al.*, 1975).

Results

The 1-hour blood-xylose concentrations are shown in Fig. 1. Only one of 46 patients with untreated coeliac disease had a value >20 mg/100 ml (mean $\pm$ SD 12 ± 5 mg/100 ml). In the control group 6 of 102 patients had 1-hour blood-xylose values <20 mg/100 ml (mean $\pm$ SD 32.8 ± 10.5 mg/100 ml). The difference between the two groups was highly significant ($P < 0.001$, t-test).

Fig. 1 One-hour blood-xylose values (mg/100 ml), giving means $\pm$ SD, in 46 children with untreated coeliac disease and 102 controls with normal jejunal mucosa.

In Fig. 2 blood xylose concentrations are shown related to the weight of the patients in the control group. As expected, concentrations fell with increasing body weight ($r = -0.6048$, $P < 0.001$).

Discussion

Despite its simplicity, the 1-hour blood-xylose value shows a surprising accuracy in separating control patients and patients with active coeliac disease, in this large series only 1 out of 46 untreated coeliac patients giving falsely normal, and only 6 out of 102 control patients falsely abnormal results. 4 of these latter 6 patients weighed above 30 kg, and so were above the upper limit of body weight recommended for the test by Rolles *et al.* (1973). In a previous investigation (Schaad *et al.*, 1975, 1977) of patients with untreated coeliac disease there was a good correlation of the 1-hour blood-xylose value with intraepithelial lymphocyte counts. It is therefore not surprising that we now find a good correlation between the morphological appearance of the mucosa and the outcome of this test.

Summary

In a series of 46 children with untreated coeliac disease and in 102 controls with normal mucosa the

1-hour blood-xylose test was, in view of its simplicity, of much value in the diagnosis of childhood coeliac disease. Only one blood-xylose result was falsely normal in the 46 coeliac patients. It is concluded that a normal blood-xylose value does not exclude coeliac disease and should not prevent peroral biopsy in the presence of strong clinical suspicion. On the other hand, patients who have repeatedly abnormal blood-xylose values merit an intestinal biopsy even in the absence of suggestive clinical symptoms.

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Fig. 2 Correlation between one-hour blood-xylose value (mg/100 ml) and body weight (kg) in 102 children with normal jejunal mucosa.

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Persistent neonatal hypoglycaemia due to glucagon deficiency

Glucagon deficiency as a possible cause of persistent neonatal hypoglycaemia has been discussed before (Wagner *et al.*, 1969; Gotlin and Silver, 1970; Zuppinger, 1975). Vidnes (1976) recently described a case of persistent hereditary neonatal hypoglycaemia due to glucagon deficiency; therapy with zinc-protamine-glucagon resulted in dramatic improvement. This was the first case in a child where glucagon deficiency was verified by glucagon determination. We here describe a further case of a newborn infant with persistent neonatal hypoglycaemia due to glucagon deficiency and treated by zinc-protamine-glucagon.

Case report

A boy was born after a normal pregnancy in August 1975 as the third child of healthy unrelated parents. He has 2 healthy siblings. No family history of infant deaths from hypoglycaemia was known. Birthweight was 3.7 kg. The child was breast fed, and the first day passed uneventfully. In the evening

of the second day he became drowsy and failed to suck. Next morning he was apathetic, breathed irregularly, and rolled his eyes. At admission to a local hospital blood glucose was low (0.9 mmol/l; 16 mg/100 ml) with a metabolic acidosis (pH 7.22, base excess -5.5 mmol/l (-5.5 mEq/l)). Treatment was started with infusion of glucose 10%; after 2 and 4 hours respectively the blood glucose value was 1.6 and 0.5 mmol/l (28.8, 9 mg/100 ml). Apart from the infusion therapy, which was continued, he received feeds of humanised milk, and prednisone was started. The first day after admission several convulsions were noted, but within a few days the clinical condition improved. During the next few weeks several attempts were made to reduce the frequency of oral feedings below 8 times per day, but each time the blood glucose level decreased and his condition deteriorated.

At the age of 3 weeks he was referred to the University Hospital. We found an alert but sweaty and restless baby with a length of 56.5 cm and a weight of 4250 g. The liver was palpable 1 cm below the costal margin. Blood was analysed for cortisol, insulin, thyroxine, growth hormone, lactate, pyruvate, and alanine levels. After 2 hours of fasting the levels of β -hydroxybutyrate and acetoacetate were respectively 37 and 15 μ mol/l (0.39, 0.15 mg/100 ml). Reducing substances in the urine were absent. Vanillylmandelic acid excretion was 4.5 nmol in 24 hours. No abnormal organic acids in the urine were detected by gas chromatography. Under a regimen of 12 oral feeds of humanised milk per day, blood glucose varied between 1.8 and 3.0 mmol/l (32, 54 mg/100 ml). A glucose tolerance test showed a high peak value after 45 minutes and rapid decrease to values below normal. Insulin regulation during the test was normal (Zuppinger, 1975; Fig. 1). A glucagon tolerance test showed a normal glucose mobilisation, followed by rapid decrease to values below normal: again insulin regulation was intact (Zuppinger, 1975; Fig. 2).

In the light of a possible glucagon deficiency we started treatment with zinc-protamine-glucagon IM at a dose of 2 mg, with 8 feeds per day. For 8–10 hours after the injection the blood glucose level remained above 3 mmol/l (54 mg/100 ml). When given twice daily, the effect lasted for 24 hours.

The clinical condition improved, and the frequency of feeds could be lowered to 6 times per day. In the next few months we had to increase the dosage of glucagon gradually, and stable glucose values could then be maintained. With a regimen of 6 feeds per day and two daily doses of 3.5 mg glucagon IM symptomatic hypoglycaemia did not occur. The patient was discharged in December 1975. After some months the parents discontinued the glucagon