Coeliac disease in Cuban children

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SUMMARY Coeliac disease is generally considered to be a disease of Europe, North America, and Australasia. A series of well-authenticated cases from Cuba is presented. One of the factors responsible for the presence of this disease in Cuba may be the increase in wheat consumption in the last few years. It is likely that coeliac disease exists in other tropical countries.

Coeliac disease has traditionally been considered as a disease of Europe, North America, and Australasia.¹ Recently, a group of cases was found in Asian children, born in England, whose parents came from the Punjab and West Pakistan.² Coeliac disease has also been reported in Sudanese children.³ Some patients have been confirmed as having coeliac disease in Mexico (L Vega Franco, 1978, personal communication; P Coello Ramirez, 1978, personal communication),⁴ but there is no further evidence of the disease in other Latin-American countries.

In Cuba some isolated patients were diagnosed as coeliacs on the basis of a good response to a gluten-free diet, but there was no confirmation by intestinal biopsy.⁵ The aim of this study was to confirm the existence of coeliac disease in Cuba, basing the diagnosis on the accepted international criteria.⁶

Subjects and methods

During the period 1972–8, 519 children from all parts of the country and aged between 3 months and 14 years, were admitted to the Instituto de Gastroenterología, either because they had a history of chronic diarrhoea with suspected malabsorption, or because they were of short stature or underweight.

In all patients a biopsy of the small intestine was obtained under fluoroscopic control with a paediatric Crosby capsule. Haematoxylin and eosin-stained sections were made, and each specimen was graded as normal, minor partial villous atrophy, major partial villous atrophy, or total villous atrophy (flat mucosa).

Chemical determination of faecal fat in 24-hour specimens collected for at least 3 days was made.⁷ A 1-hour blood xylose test was used to investigate intestinal absorption, and a level <20 mg/100 ml was considered as indicative of malabsorption.^{8–9}

Duodenal juice and faeces were examined for parasites.

Fifty patients each had a flat mucosa to some extent, together with clinical or biochemical evidence of malabsorption. Each was treated with a gluten-free diet. The details of the gluten-free diet were explained to the patients, or to the parents of the patients, by a dietitian.

Each child suspected of being a coeliac was followed-up periodically at a special clinic where emphasis was placed on progress in weight and height, and on the relief of symptoms after withdrawal of gluten from the child's diet (Fig. 1).

After a year of treatment a second biopsy (Fig. 2) was taken, and a xylose test made.

Twelve of these 50 children were readmitted and each received a dose of 30 g gluten a day for 3 days, and then a third biopsy was taken (Fig. 2) and a further xylose test done. In these patients the villous height of 100 epithelial cells was measured with an eyepiece micrometer after gluten exclusion, and again after the reintroduction of gluten.

Results

Fifty (8.8%) patients were diagnosed as possible coeliacs (Figs 1, 2). Five patients were diagnosed in 1972, 8 in 1973, 7 in 1974, 9 in 1975, 13 in 1976, 7 in 1977, and 1 in 1978. Thirty (60%) of them were girls. The remaining patients had giardiasis, lactose intolerance, irritable bowel syndrome, etc. None had tropical sprue.

Forty-two (84%) patients were white, 2 (4%) were black, and 6 (12%) were mestizos (2 of these 6 were of Asian origin). The parents of one patient came from Chile and the ancestors of another patient were black, immigrants from the Caribbean island of Grenada.

Failure to thrive was the predominant way in which coeliac disease presented, especially in those

Fig. 1 Cuban girl with coeliac disease (a) before, and (b) 3 months after gluten-free diet.

over 5 years of age. Those who were diagnosed in early childhood had generally been referred with a history of chronic diarrhoea.

The results of the faecal fat and the xylose tests were that 24 (48%) patients had steatorrhoea, and 40 (80%) had abnormal results for the xylose test.

The Table shows the morphological grading of the intestinal biopsy, before and after a gluten-free diet. A second biopsy was omitted in 10 children because they had not spent sufficient time on a gluten-free diet.

Of the 12 patients in whom a gluten challenge was made, there was a fall in the blood level of xylose in 4. In the biopsies, the mean cell height after the gluten-free diet was $28.9 \mu\text{m}$, with a range of 25–33. After the reintroduction of gluten, the mean cell

Table Small intestine biopsy results in 50 children with possible coeliac disease, before and after gluten-free diet

Morphological grading	At initial diagnosis		After gluten-free diet*	
	No	%	No	%
Normal	—		10	25
Minor partial villous atrophy	6	12	16	40
Major partial villous atrophy	13	26	8	20
Total villous atrophy	31	62	6	15
Total	50		40	

*10 cases were not rebiopsied (see text).

height fell to $26.1 \mu\text{m}$, with a range of 16–34. With regard to the villous height after the gluten-free diet, the mean value was $253 \mu\text{m}$; after the gluten challenge, the mean villous height fell to $221 \mu\text{m}$.

Fig. 2 Biopsies of same girl as Fig. 1 (a) at initial diagnosis, (b) after gluten-free diet, (c) after gluten challenge.

Discussion

The criteria for the diagnosis of coeliac disease have been discussed in several forums, and are currently based on a series of changes of the intestinal mucosa in relation to gluten elimination and challenges. When we realised that coeliac disease existed not infrequently in Cuba, we accepted the necessity to adhere to the basic principles for the diagnosis of coeliac disease. Nevertheless, the strict criteria cannot always be applied¹⁰ and some paediatricians consider that the diagnosis of coeliac disease can be established with the demonstration of a clinical and histological response to a gluten-free diet. At the same time, although a flat intestinal mucosa is characteristic of the disease, and in those countries where coeliac disease is present the majority of children found to have a flat intestinal mucosa do have coeliac disease, other reasons for such a mucosal pattern have been known to exist. Transient gluten intolerance has been described¹¹ and should be considered as part of the differential diagnosis with coeliac disease.

Of our 50 cases of presumed coeliac disease only 12 children fulfilled strict diagnostic criteria. Long-term follow-up and subsequent reinvestigation of the other 38 children with flat mucosae and clinical and histological responses to gluten withdrawal should enable us to confirm the diagnosis.

Coeliac disease is mainly a disease of Europe, North America, and Australasia; no convincing case has been recorded in China. It had been thought that the disease was hardly ever found in people of negro origin and we therefore stress that such children were among our cases.

The factors that may contribute to the racial and geographical incidence of coeliac disease are genetic predisposition and diet.¹² We have no information about the presence in the Cuban population of HL-A8, considered to be a predominately Caucasian antigen. The incidence of coeliac disease among whites in our cases appears high, but the wheat consumption of our population has greatly increased in the last few years.

Coeliac disease may be more common in tropical countries than had been appreciated. Paediatricians of developing countries should differentiate the clinical picture of these young infants from enteric infections or other causes of diarrhoea, in order to treat this disease correctly and prevent complications—such as severe growth retardation and delayed development.

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