

Coeliac disease: a potentially treatable health problem of Saharawi refugee children

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Objective To characterize the clinical and nutritional impact of coeliac disease (gluten-sensitive enteropathy) among Saharawi children living as refugees in Algeria.

Methods A total of 65 Saharawi children with coeliac disease were compared with 71 age-matched non-coeliac controls. For each participant, the clinical history was taken and a clinical examination, non-quantitative 24-hour dietary recall, anthropometric and skinfold measurements, bioelectric impedance analysis (BIA) of body composition, and venous blood sampling for haemoglobin determination were performed.

Results Gluten-containing food, especially bread, was the staple diet of Saharawi children. Abdominal pain and distension were significantly commoner among children with coeliac disease than in controls ($P < 0.05$). The mean height-for-age was significantly lower in such children than in controls (-2.5 ± 1.4 units vs -1.8 ± 1.3 units, respectively, $P < 0.01$). No significant differences were found for either skinfold or BIA measurements. Haemoglobin values tended to be lower in children with coeliac disease than in controls.

Conclusions Coeliac disease has a negative effect on the health status of Saharawi refugee children. Because of the high prevalence of the condition in the Saharawi, a specific programme for treating all affected individuals should be established. Further studies are required to quantify the impact of coeliac disease in other areas of the developing world.

Keywords Celiac disease/ethnology; Diet/adverse effects; Health status; Refugees; Child; Africa, Northern; Algeria (*source: MeSH*).

Mots clés Coeliaque, Maladie/ethnologie; Régime alimentaire/effets indésirables; Etat sanitaire; Réfugié; Enfant; Afrique nord; Algérie (*source: INSERM*).

Palabras clave Enfermedad celíaca/etnología; Dieta/efectos adversos; Estado de salud; Refugiados; Niño; África del Norte; Argelia (*fuelle: BIREME*).

Bulletin of the World Health Organization, 2001, **79**: 541–545.

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Introduction

Individuals who suffer from coeliac disease have a permanent intolerance of dietary gluten, a protein occurring in wheat, rye, and barley. The condition, which is characterized by severe, immune-mediated damage to the jejunal mucosa (subtotal villous atrophy), typically involves chronic diarrhoea, abdominal distension, weight loss, and malnutrition. Diagnosis is based on the detection of specific circulating antibodies, especially class A endomysial and anti-transglutaminase antibodies, and on typical enteropathy upon small intestinal biopsy. Treatment with a gluten-free diet results in healing of the enteropathy and normalization of the clinical picture (1, 2).

Coeliac disease is a common disorder among the Saharawis, a population that originally lived in the

Western Sahara in both settled and nomadic communities. Following political changes during the 1970s, approximately 150 000 Saharawis left their territory and settled in a desert area near Tindouf, Algeria, where they remain encamped. In a sample of 989 Saharawi refugee children we recently reported a 5.6% prevalence of coeliac disease (3), five to ten times the frequency in developed countries, where the prevalence lies in the range 0.5–1% (4, 5). The consequences of coeliac disease for the health status of Saharawi refugee children are not clear, partly because complaints that are characteristic of the disease can be caused by infectious diarrhoeal diseases and primary malnutrition, which occur frequently in this population (6). We therefore studied the clinical and nutritional impact of this chronic disorder in a group of 65 Saharawi children, who were compared with an age-matched group of controls free of the disease.

Materials and methods

The patients were 65 Saharawi refugee children with active coeliac disease who were diagnosed between

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January 1998 and February 1999. The diagnosis was based on detection of class A endomysial antibodies by indirect immunofluorescence assay of serum at 1:5 and 1:50 dilutions, with monkey oesophagus as the antigenic substrate (Antiendomysio Eurospital, Trieste, Italy). For a subset of 16 patients the serological diagnosis was confirmed by finding the typical coeliac enteropathy upon intestinal biopsy (3). There were 38 female and 27 male patients (mean age \pm standard deviation (SD) = 7.7 ± 3.0 years; range

= 1.5–15.5 years), including five sibling pairs. At the time of the study the patients' carers were not aware of the diagnosis and the related dietary recommendations. Controls were 36 female and 35 male Saharawi children (mean age \pm SD = 7.0 ± 3.7 years; range = 1.1–16 years), who were randomly selected from the apparently healthy paediatric population and were negative when tested for serum class A endomysial antibodies.

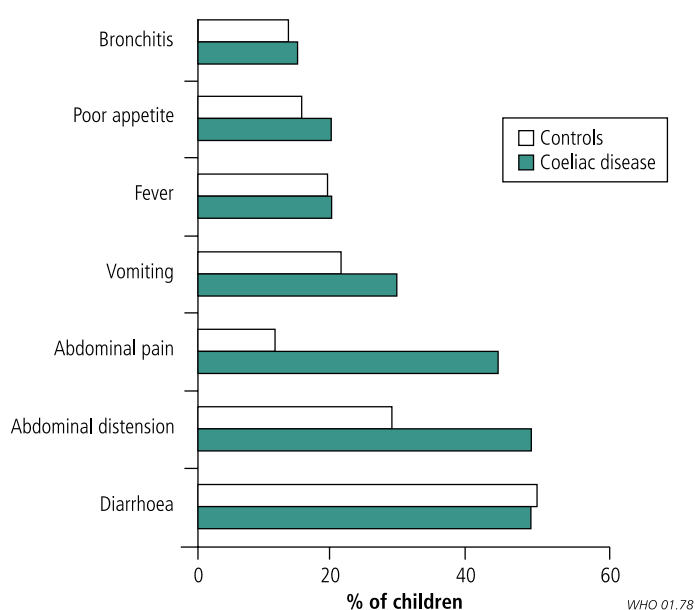
The following were conducted for each study subject, with the informed consent of the carer: clinical history and examination; non-quantitative 24-h dietary recall and nutritional history of the early years of life (duration of breastfeeding, age at weaning, first solid food); anthropometric measurements (weight, height, triceps and subscapular skinfold measurements); bioelectric impedance analysis (BIA) of body composition using an Akern BIA 101 analyser (Akern, Florence, Italy) with an operating frequency of 50 kHz at 800 μ A and standard electrode locations on the right hand and foot (the body's resistance and reactance plus height, weight and age were used in equations provided by the manufacturer to obtain an estimate of fat-free mass and fat mass); and venous blood sampling for haemoglobin determination.

Graphpad Prism software was used for statistical analysis and graphics.

Table 1. The most common foods ingested by children during the 24 hours before being examined

Item	Frequency of consumption (%)
1. Bread	88
2. Rice	63
3. Potatoes	27
4. Meat	27
5. Human milk	19
6. Cheese	19
7. Couscous	15
8. Oil	15
9. Milk	15
10. Biscuits	10
11. Tea with sugar	10
12. Lentils	10
13. Onions	10
14. Jam	7
15. Eggs	7
16. Dates	5
17. Pasta	2
18. Sardines	2

Fig. 1. Prevalence of symptoms among study children during the 8 weeks prior to examination. Abdominal pain and distension were significantly commoner among children with coeliac disease ($P < 0.05$).



Results

Table 1 shows the cumulative frequency of food items ingested during the 24 h before examination by the children with coeliac disease and by the controls, there being no difference between the two groups in this respect. Gluten-containing food, especially bread, was the staple item in the children's diet. The mothers of 23 of the younger children were able to recall the duration of breastfeeding \pm SD (18 ± 7 months; range = 2–36 months) and the age at weaning \pm SD (15 ± 8 months; range = 8–36 months). Bread was usually the first food introduced at weaning.

Fig. 1 and Fig. 2 show, respectively, the prevalence of symptoms among study subjects during the 8 weeks prior to interview and their mean anthropometric indices (Z-scores). The proportions of study patients and controls, respectively, who had values for these indices less than -2 SD of the mean were as follows: 63% and 49% for height-for-age; 63% and 54% for weight-for-age; and 17% and 31% for weight-for-height. No significant differences were found in either skinfold or BIA measurements between subjects with coeliac disease and controls. The respective BIA measurements for fat-free mass were $74.9 \pm 12.3\%$ and $75.9 \pm 9.4\%$, and those for fat mass were $25.8 \pm 12.4\%$ and $24.5 \pm 9.5\%$.

Haemoglobin values tended to be lower in children with coeliac disease than in control subjects (Fig. 3)

Discussion

The reasons for the unusually high frequency of coeliac disease among the Saharawi people are probably related to both genetic and environmental factors. Previous studies have suggested that gluten-sensitive enteropathy could be a common disorder in North African and Eastern Mediterranean populations (7–9). An HLA analysis of 94 Tunisian children with coeliac disease showed that all of them carried at least one of two synergistic heterodimers that predispose to the condition: a DQ heterodimer, encoded by *DQA1*0501* and *DQB1*0201* and/or a DR heterodimer, encoded by the non-polymorphic gene *DRA* and the *DRB4* gene (10). The Saharawi are a highly inbred population of Arab and Berber origin. Preliminary data suggest a strong association of the *DR3*, *DQA1*0501-DQB1*0201*-positive haplotypes and genotypes with coeliac disease in these Saharawi children, at a similar level to that observed in other populations (F. Cucca, unpublished data, 2001).

On the other hand, nutritional factors also have a bearing on the pathophysiology of coeliac disease. Historically, the Bedouin diet was based on prolonged breastfeeding, camel milk and meat, dates, sugar, and small amounts of cereals and legumes. Over the last century, however, the Saharawi dietary habits have changed dramatically, and products made with wheat flour, especially bread, have become the staple food. Gluten-containing cereals, especially wheat, can provide more than 50% of total dietary energy in North African countries (11). Among the Saharawi the duration of exclusive breastfeeding has progressively reduced, and bread is often introduced into the infant's diet during the first year of life. Such abrupt dietary changes could have accounted for the large number of cases of coeliac disease observed in Saharawi children.

The predominant clinical picture of coeliac disease among Saharawi children usually involves chronic diarrhoea, abdominal distension, growth failure, depressed mood, and loss of appetite. The risks of developing severe diarrhoea and of dying from dehydration are greatest among the youngest children, especially during summer. Height-for-age values reflect linear growth, and if they are too low, i.e. if stunting occurs, long-term cumulative inadequacies in health and/or nutrition are indicated. Among Saharawi children the prevalence of stunting is very high (> 40%) in terms of the WHO classification (12). It was recently reported that the mean height-for-age was -1.9 ± 1.6 SD of that of the NCHS/WHO reference population among 424 children under 5 years of age and -1.4 ± 1.2 SD among 512 children aged 5–10 years, with prevalences of stunting (height-for-age < -2 SD of the NCHS/WHO reference population) of 46.4% and 30.7%, respectively (6). The mean height-for-age (-1.8 ± 1.3 SD of the NCHS/WHO reference population) and the high prevalence of stunting (49%) in our control group were similar to these values and probably arose from a combination of

Fig. 2. Mean anthropometric indices of study children (one-sided bars show standard errors). Height-for-age was significantly lower in children with coeliac disease than in controls (-2.5 ± 1.4 units versus -1.8 ± 1.3 units, respectively; $P < 0.01$)

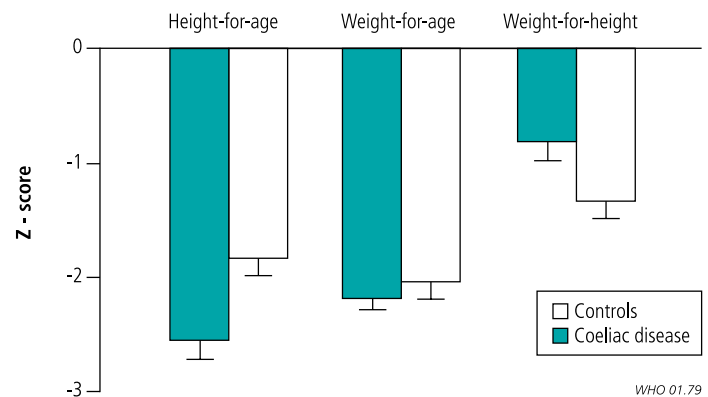
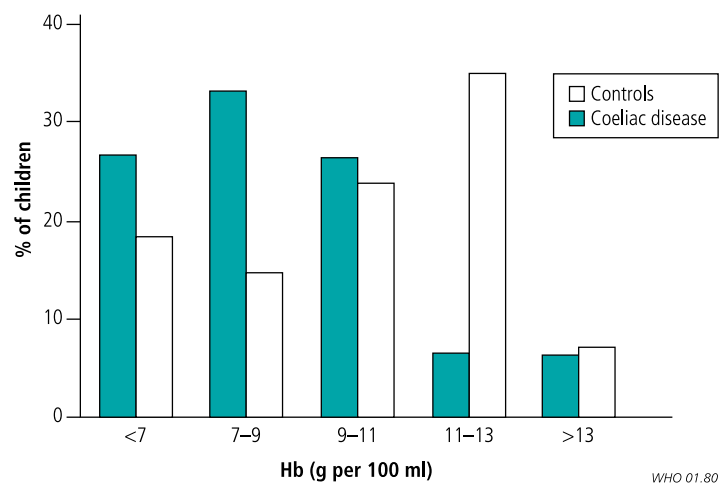


Fig. 3. Distribution of haemoglobin (Hb) levels in the study children



poor nutrition and the impairment of nutrient absorption by intestinal infections and infestations. In Saharawi children with coeliac disease the degree of stunting was even more pronounced, with a mean height-for-age of -2.5 ± 1.4 SD of the NCHS/WHO reference population, significantly lower than the control value, and with 63% of children having values < -2 SD. This could arise because of chronic impairment of intestinal absorption resulting from the decreased absorptive area, impaired enzyme activity on the brush border of the enterocytes, secondary pancreatic insufficiency, and the exudation of proteins into the gut (1). In developing countries, severe stunting is associated with an increased risk of mortality (13), especially among children with protracted diarrhoea (14).

Anaemia is a common health problem among the general Saharawi population, especially children and women. It is usually related to several factors, such as poor iron and vitamin C intake and increased blood loss in the gut caused by chronic infestations. In the study, children with coeliac disease tended to have even lower haemoglobin levels than the controls (Fig. 3). This is not surprising, since iron

absorption, which normally takes place in the duodenum and the first portion of the jejunum, is greatly impaired by coeliac enteropathy. Generally, in cases of coeliac disease, iron deficiency cannot be corrected by oral iron supplementation in either food or pharmacological preparations. However, iron absorption reverts to normal after initiation of a gluten-free diet (15).

Treatment of coeliac disease is based on lifelong exclusion from the diet of gluten-containing cereals, i.e. wheat, barley, and rye. In most developed countries this is easily accomplished by using both cereals that do not contain gluten (e.g. rice and maize) and palatable gluten-free, commercially available products that are specifically designed for patients with coeliac disease. In contrast, treating the disease in the Saharawi context is exceptionally difficult. The living conditions of the refugees are harsh, with severe problems of water and food supply. In addition, the general ration covers only 68% of the estimated energy requirement of the population (6). We are currently developing a multifaceted project with the aim of improving the treatment of coeliac disease in Saharawi children. To be effective, implementation of a gluten-free diet has to take local dietary habits into account. Rice and millet, which are locally available, are gluten free (16) and could be the staple starchy food in the diet of Saharawis with coeliac disease; however, in order to avoid cross-contamination with gluten, dedicated machinery needs to be used to mill these cereals (17). Since the diarrhoea associated with coeliac disease can be partly linked to secondary lactase deficiency, yoghurt, instead of milk, can be used during the first months of treatment (18). Our treatment strategy also includes the following elements: educational courses for

doctors, nurses, dieticians, school personnel, affected families, and the general population; identification of all affected individuals in a register; centralized management of gluten-free food storage and distribution; and setting up a group to help patients with coeliac disease to cope with the treatment and maintain contacts with other national societies and international agencies.

The present study shows that coeliac disease has a negative effect on the health status of Saharawi refugee children. Because of the high prevalence of this condition among the Saharawi people, a specific programme for treating all affected individuals should be established. Further studies are required to quantify the impact of coeliac disease in other areas of the developing world. ■

Acknowledgements

We thank the Saharawi Health Authorities for authorizing this study and for logistical support. The work in the Saharawi camps was greatly facilitated by the tireless efforts of Mr Maana El Ouali Bahia, a laboratory technician at the Rabuni Hospital, and Mrs Patrizia Menegazzi of the Department of Paediatrics, Livorno Hospital. The work was supported by grants from Cooperazione per lo Sviluppo dei Paesi Emergenti, Florence, Italy, and Associazione Italiana di Solidarietà con il Popolo Saharawi. Special thanks go to Professor Luciano Vizzoni, Department of Paediatrics, Livorno Hospital, and Dr Sandro Guiglia, Cooperazione per lo Sviluppo dei Paesi Emergenti, for their assistance in managing the project.

Conflicts of interest: none declared.

Résumé

Maladie cœliaque : un problème de santé potentiellement curable chez les enfants sahraouis réfugiés

Objectif Caractériser l'impact clinique et nutritionnel de la maladie cœliaque (entéropathie due à une intolérance au gluten) chez les enfants sahraouis réfugiés en Algérie.

Méthodes Nous avons comparé 65 enfants sahraouis atteints de maladie cœliaque avec 71 témoins appariés indemnes de cette affection. Pour chaque participant, les antécédents cliniques ont été recueillis et il a été procédé à un examen clinique, un rappel non quantitatif de l'alimentation sur les précédentes 24 heures, des mesures anthropométriques et la mesure du pli cutané, une analyse de la composition corporelle par impédancemétrie (BIA) et un prélèvement de sang veineux pour la mesure de la concentration en hémoglobine.

Résultats L'alimentation de base des enfants sahraouis était constituée d'aliments contenant du gluten, et en particulier de pain. Les douleurs abdominales et le ballonnement étaient significativement plus fréquents

chez les enfants atteints de maladie cœliaque que chez les témoins ($p < 0,05$). Le rapport taille/âge moyen était significativement plus faible chez ces enfants que chez les témoins ($-2,5 \pm 1,4$ unités contre $-1,8 \pm 1,3$ unités, $p < 0,01$). Aucune différence significative n'a été observée au niveau du pli cutané ni des mesures impédancemétriques. Les taux d'hémoglobine tendaient à être plus faibles chez les enfants atteints de maladie cœliaque que chez les témoins.

Conclusion La maladie cœliaque a un effet négatif sur l'état de santé des enfants sahraouis réfugiés. Du fait de la forte prévalence de cette affection chez les Sahraouis, un programme spécifique de traitement de toutes les personnes atteintes devra être établi. D'autres études sont nécessaires pour chiffrer l'impact de la maladie cœliaque dans d'autres régions du monde en développement.

Resumen

Enfermedad celíaca: un problema de salud potencialmente tratable de los niños saharauis refugiados

Objetivo Caracterizar la repercusión clínica y nutricional de la enfermedad celíaca (enteropatía por sensibilidad al gluten) entre niños saharauis que vivían como refugiados en Argelia.

Métodos Se procedió a comparar a 65 niños saharauis con enfermedad celíaca con 71 testigos de edad similar no afectados por la enfermedad. En cada caso se llevó a cabo lo siguiente: historia clínica, exploración física, anamnesis nutricional no cuantitativa de 24 horas, mediciones antropométricas y del pliegue cutáneo, análisis de la composición corporal mediante impedancia bioeléctrica, y toma de muestras de sangre venosa para determinar la hemoglobina.

Resultados La alimentación de base de los niños saharauis eran productos que contenían gluten, especialmente pan. La presencia de dolor y distensión abdominales fue significativamente más frecuente entre

los niños con enfermedad celíaca que entre los testigos ($P < 0,05$). El valor medio de la estatura para la edad fue significativamente más bajo en esos niños que en los testigos ($-2,5 \pm 1,4$ unidades frente a $-1,8 \pm 1,3$ unidades, respectivamente, $P < 0,01$). No se detectaron diferencias significativas en lo relativo al pliegue cutáneo o la impedancia bioeléctrica. Los niveles de hemoglobina tendían a ser menores en los niños con enfermedad celíaca que en los testigos.

Conclusión La enfermedad celíaca repercute negativamente en el estado de salud de los niños refugiados saharauis. Dada la alta prevalencia de esa dolencia en la población saharauí, debería establecerse un programa específico para tratar a todos los individuos afectados. Es necesario realizar nuevos estudios para cuantificar el impacto de la enfermedad celíaca en otras regiones del mundo en desarrollo.

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