

Compliance in teenagers with coeliac disease—a Swedish follow-up study

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Ljungman G, Myrdal U. Compliance in teenagers with coeliac disease—a Swedish follow-up study. *Acta Paediatr* 1993;82:235–8. Stockholm. ISSN 0803–5253

A group of 47 children with coeliac disease, born between 1973 and 1978 in the Swedish county of Västmanland, participated in a controlled questionnaire study. Health, self-esteem, knowledge of the disease and dietary compliance were investigated. The children with coeliac disease were otherwise just as healthy as the control children. Growth and self-esteem were normal. Good knowledge of coeliac disease and dietary treatment was found in 87% of children and dietary compliance was 81%. Girls and younger children (12–14 years) were more compliant than boys and older children (15–17 years). Compliance correlated positively to knowledge. □ *Coeliac disease, dietary compliance, health, teenagers*

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Many studies have been carried out on different aspects of childhood coeliac disease (CD). However, little has been done on how these children experience their situation and how they cope with dietary treatment. Two British studies (1, 2), an Italian study (3) and a Finish study (4) have dealt with these questions. In both British studies, dietary compliance was approximately 60%, in the Italian study it was 65% and in the Finish study it was 76%. Treatment of CD is a strict gluten-free diet (GFD) for life. This is based on the knowledge that when gluten is reintroduced, sooner or later small bowel mucosal pathology recurs, even though symptoms may be absent (5–7). Furthermore, Holmes et al. (8) have suggested that lifelong dietary treatment may have a protective role against development of malignancy.

The aim of this study was to assess health, self-esteem, dietary compliance and knowledge of the disease and its treatment in a Swedish group of teenagers with CD more than 10 years after diagnosis.

Patients and methods

Västmanland is a county in the central part of Sweden with 255 000 inhabitants. The county has one major city of 120 000 inhabitants, Västerås, and three smaller towns. The main occupations are found in industry, trade and public services. The social structure in the county is comparatively homogenous with low unemployment figures, with all children attending nine years of compulsory school free of charge. In the area, 19 311 children were born between 1973 and 1978. Ninety-eight percent of children are regularly followed-up at child health centres. Trained nurses and paediatricians record weight and height on growth charts at least six times

during the first year of life in order to detect signs of malnutrition or other disorders. During the study period, formulas containing gluten were introduced at four months of age in most children. Children and adolescents with CD are prescribed gluten-free items with a large discount to eliminate additional costs to the family. A special gluten-free menu is offered in all school canteens.

All children with suspicion of CD were referred to the only paediatric clinic in the county for intestinal biopsy and were registered. The diagnosis of CD was based on the 1969 ESPGAN criteria (9) with three biopsies and clinical response to GFD. Forty-seven children born between 1973 and 1978 fulfilled these criteria: these patients became the study group and all of them were located. Nine of the 47 children had moved out of the county. For practical reasons, two controls for each CD patient still living in the county were chosen. This resulted in a control group of 76 children matched for age, sex, school class and geographical distribution.

All the above mentioned patients with CD were diagnosed before two years of age. After the study had started, a girl born in 1977 was found to have CD at 15 years of age. The prevalence of CD in this population is 2.59/1000 and the sex ratio boy/girl is 1/2.8. All children with CD are regularly followed-up until 20 years of age by paediatricians associated with the clinic.

A two-part mailed questionnaire was used. The first part, was intended to obtain a picture of health and self-esteem and was distributed both to the study group and to the control group. Most of the questions in this part have been used in other studies and validity has been tested and proved to be good (10). The second part, evaluating dietary compliance and knowledge of the disease and its treatment, was sent only to the coeliac

Table 1. Frequency (%) of other diseases in patients with coeliac disease (CD) and in control subjects.

	CD group (n=47) (%)	Control group (n=66) (%)	<i>p</i>
Diabetes mellitus	4	2	ns
Allergic rhinoconjunctivitis	9	15	ns
Asthma	2	5	ns
Eczema	13	20	ns
Cow's milk allergy	0	2	ns
Thyroid gland disease	0	0	ns
Joint disease	0	3	ns

Table 2. Frequency of symptoms in coeliac disease (CD) and control groups.

	CD group (n=47) (%)	Control group (n=66) (%)	<i>p</i>
Nausea	19	20	ns
Diarrhoea	23	11	*
Constipation	4	5	ns
Abdominal pain	34	42	ns
Headache	68	64	ns
Neck pain	19	23	ns
Back pain	27	26	ns
Leg pain	15	20	ns
Sleep disorders	27	11	*
Downheartedness	15	18	ns
Difficulties in relaxing	28	13	*
Tiredness	36	46	ns
Poor appetite	15	16	ns

* $p < 0.05$.

group: there were 15 questions testing knowledge. Knowledge was considered to be good if more than 12 answers were correct. The children were instructed to answer the forms together with their parents, but we stressed that the child's own responses to the questions were those required. All 47 children in the CD group replied while 10 of the 76 in the control group did not. Almost all questions were answered.

Chi-square test and Student's *t*-test were used for statistical analysis. Critical value for statistical significance was $p < 0.05$.

Results

Health and self-esteem

Mean height was the same for children with CD and the controls (169.1 cm = +0.61 SD) and there was no significant difference in body weight between the groups

Table 3. Frequency of gluten intake by mistake or neglect in patients with coeliac disease at time of study.

	%	<i>n</i>
Gluten free diet (gluten less than once a month)	81	38
Moderate dietary neglect (gluten more than once a month)	11	5
Clear dietary neglect (gluten more than once a week)	2	1
Total dietary neglect—normal diet (gluten every day)	6	3
Total	100	47

Table 4. Compliance to the gluten free diet in various situations in coeliac disease (CD) group and in subgroups.

	Total (n=47) (%)	Girls (%)	Boys (%)	12–14 yr (Younger) (%)	15–17 yr (Older) (%)
At home	92	94	83	96	86
In school	87	91	75	96	77
Out with friends	77	82	58	87	64

(52.4 kg = +0.11 SD) versus (53.3 kg = +0.22 SD). Separate analysis comparing compliers, non-compliers and controls showed no significant differences or even tendencies concerning height or weight. One of the three totally non-compliant children who had been on a gluten-containing diet since two years of age was short at the time of the study (159 cm = -2.1 SD at 15 years of age).

In the CD group, 44% had been in contact with some form of medical care, including school nurses, during the past year. The corresponding figure for the control group was 57%. The girls in both groups had more medical contacts than the boys (56% versus 34%). In the questionnaire there was a large number of questions concerning different diseases and symptoms. Two of the children with CD (4.2%) and one of the controls (1.5%) had diabetes mellitus. Diarrhoea, sleep disorders and difficulties in relaxing were significantly more common in the CD group. Other significant differences were not demonstrable, but allergic disorders, abdominal pain and tiredness were more common in the control group (Tables 1 and 2).

There were no differences between the groups in their estimation of health, physical constitution, self-esteem or fitness. No differences in attitudes to school, school meals, sports, classmates or perception of health were noted. Both groups saw friends to the same extent during leisure time.

Knowledge about CD

Eighty-seven percent of the children had correctly answered more than 12 of the 15 questions on CD or diet, and thus by our definition had good knowledge. The remaining 13% less often knew someone else with CD (40% versus 67% in the well informed group), were less often members of the Coeliac Society (20% versus 44%) and had check-ups less often. These six patients were also more discontented with their doctor and their medical care on the whole (40% versus 2%). The children with good knowledge considered themselves to be well informed, while the others did not.

Dietary compliance

Among the CD children, 22 stated that they never ate any food containing gluten and 25 stated that they sometimes did. Of those eating food containing gluten, 16 stated that they did so less than once a month and that it was by mistake. If we accept this as a GFD, the compliance rate is 81% (Table 3). As expected, compliance diminished when the children were not at home (Table 4). Dietary compliance was higher in girls than in boys. Compliance was also higher in younger children (12–14 years) compared to older children (15–17 years).

In the complying group, a mean of 14.03 of 15 questions testing knowledge were answered correctly versus 12.44 in the non-complying group. In the complying group, 95% considered themselves to be well or reasonably well informed versus 67% in the non-complying group. Thus knowledge of CD and the feeling of being well informed correlated significantly with compliance in the study group. The neglectors were found to have poor knowledge.

The GFD was experienced as troublesome by 77% of patients while 17% denied this. The remaining 6% were the children not complying with the GFD. Those who found it troublesome described the troubles as moderate.

Discussion

Colaco et al. have reported a negative effect on growth in adolescents not adhering to a GFD (11). We did not find any noticeable differences in height or weight. Possible explanations for this could be that the non-compliers were a small group and that they may have complied better during more sensitive periods of growth.

The adolescents with CD were as healthy as the controls. A connection between CD and diabetes mellitus has previously been described (12). In the study group, there are two CD patients with diabetes mellitus but the numbers were too small to analyse statistically.

Measuring self-esteem is always a delicate task. We tried to do this by allowing the adolescents to estimate a large number of different indirect parameters and by

allowing them to estimate their self-esteem directly. We found no differences between the study group and the control group.

Compliance with a GFD in this population was 81%, which is higher than that found by others (1–3) and comparable with that found by Mäki et al. (4). Six percent of patients were totally non-compliant, which is in accordance with the above mentioned British studies. Mayer et al. reported 23.6% on a normal diet (3). Like Jackson et al. (1) we also found a correlation between knowledge and compliance. Older children were less strict in their diet. One explanation for this is that older teenagers more often eat snacks containing gluten while with friends and visiting restaurants. Another explanation could be that some doctors tend to accept some deviation from strict GFD after puberty when the most important period of growth and development has passed. Girls were adhering to the GFD better than boys. Part of this difference was due to the fact that there were relatively more girls in the younger group, where compliance was better.

CD is a very common disease in Sweden today and in most schools there are children who are served a GFD. Awareness and knowledge of CD is high among parents and health care personnel. This contributes to the high detection rate and also to a tolerant atmosphere where a child with CD does not feel very different.

This can be considered to be an inclusive study of CD patients born in the county from 1973 to 1978. The few drop-outs and the fact that almost all questions were answered contributed to better validity and reliability.

In conclusion, our current practice of medical care for children with CD appears to be successful and appropriate in our Swedish society, with a high frequency of the disease. We must be attentive to signals of lack of knowledge and poor compliance and be aware of the fact that compliance decreases with age. New methods of spreading knowledge and increasing motivation may be worth evaluating. Education and discussion groups are methods used in diabetes care (13) and we plan to apply these in the care of CD. Making gluten-free meals available in restaurants could also be a way of increasing compliance. It is interesting to note that in Sweden one international fast food chain took the initiative to introduce a gluten-free hamburger meal in 1991.

Acknowledgements.—The authors thank Stefan Sörensen, PhD, Department for Research, JAMU, University of Uppsala, Central Hospital, Västerås, Sweden, for support in statistical and epidemiological matters.

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Received June 22, 1992. Accepted Oct. 8, 1992

Acta Pædiatr 82: 238. 1993

LETTER TO THE EDITOR

Nasal colonization with coronavirus and apnea of the premature newborn

Sir,

Coronaviruses are a group of RNA-containing agents, isolated in 1965 (1), responsible for mild upper respiratory tract infections in adults and children (2, 3). Their pathogenicity in newborns is not well documented; enterocolitis in neonates and lower respiratory tract infections in young children have been reported (4, 5).

From March to October 1991, we observed four cases of apneic spells in hospitalized premature newborns (mean gestational age, birth weight and age at diagnosis, respectively: 30 ± 1 weeks, 1420 ± 330 g, 19 ± 8 days). Study of their nasal secretions by indirect immunofluorescence was positive with an anti-coronavirus monoclonal antibody (Biosoft-Clonatec) and negative for adenovirus, respiratory syncytial virus, myxovirus influenza and parainfluenza. These apneic spells were associated with bradycardia. They occurred unexpectedly and had no further explanation (patent ductus arteriosus, bacterial infection, central nervous system damage). Electroencephalogram failed to prove subclinical seizures. Apneas were resistant to methylxanthine and gastroesophageal reflux treatment. Neither cough nor rhinorrhea was observed. Chest X-ray remained unchanged. All neonates recovered within 10 days. In two cases, subcutaneous atropine 0.01 mg/kg reduced bradycardia. In two cases, assisted ventilation was necessary.

Apneas are frequent in premature infants. Some are associated with viral diseases, mainly respiratory syn-

cytial virus infection, occurring in outbreaks in neonatal intensive care units (6). The role of coronaviruses in nosocomial respiratory illness in neonates may be underestimated because coronaviruses are not systematically searched for. Culture is difficult but tests using monoclonal antibodies or specific nucleic probes should give further evidence of their presence. We are conducting a prospective study to determine precisely the real incidence and risk factors of coronavirus-related nosocomial infections in neonates.

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