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Evaluation of hearing loss in pediatric celiac patients

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ABSTRACT

Objectives: Celiac disease (CD) is a chronic immune-mediated enteropathy caused by the ingestion of gluten in genetically predisposed individuals. In some reports, sensorineural hearing loss (SNHL) has been identified as an extraintestinal symptom of CD. We aimed to further investigate the possible association between CD and SNHL by examining a greater number of pediatric CD patients.
Methods: The study was carried out from March to September 2014 and included 110 pediatric patients with biopsy-confirmed CD (220 ears) and 41 age- and sex-matched controls (82 ears); participants were evaluated by tympanometry and pure tone audiometry (frequency, 250–8000 Hz frequency).
Results: Audiometric bone conduction thresholds were significantly different between the CD patients and the controls ($p < 0.05$), but there were no significant differences in pure tone averages for air conduction ($p > 0.05$). When the results for CD patients were analyzed according to duration of disease ($\leq$36 months and >36 months), a significant difference in bone conduction thresholds ($p < 0.05$) was noted, with significant increments at the later stages of disease. However, this difference was not sufficient to define clinical hearing loss, as the pure tone average thresholds remained below 20 dB.
Conclusion: These results indicate that subclinical hearing loss may be present in children with CD, which could presage more serious hearing impairments at older ages and later stages of the disease. Hearing screenings should be recommended for children with CD in order to prevent the potentially unfavorable effects of hearing loss on the emotional, behavioral, cognitive, and sensorimotor development of these patients.

1. Introduction

Celiac disease (CD) is a chronic immune-mediated enteropathy caused by the ingestion of gluten in genetically predisposed individuals [1]. A CD prevalence between 0.5 and 1% has been reported in various parts of the world, including the United States, Europe, Australia, North Africa, the Middle East, India, and parts of northern China [2].

In patients with CD, gluten directly stimulates the production of specific anti-tissue transglutaminase antibodies in the small intestine as well as in the extraintestinal tissues [3]. Intestinal symptoms are common in the first year of life, while extraintestinal symptoms are more commonly reported in late childhood [4]. By adulthood, neurological symptoms may be seen in up to 36% of patients [5] and the incidence of peripheral neuropathy in this population can be up to 50% [6].

The relationship between sensorineural hearing loss (SNHL) and autoimmune diseases including Sjögren's syndrome, systemic lupus erythematosus, and inflammatory bowel diseases, has been well described [7–9]. A recent study [10] has suggested that SNHL should also be regarded as a neurological symptom of CD, and the prevalence of subclinical SNHL in association with CD has been reported to be 8.5–47.1% [4,10–13]. While there are a few published studies that assess the presence of SNHL among CD patients, the reliability of these studies has been controversial because of the small number of patients.

In the present study, we aimed to further investigate the association between CD and SNHL by examining a greater number of pediatric CD patients.

2. Materials and methods

From March to September 2014, 110 patients (220 ears) of the Clinics of Pediatric Gastroenterology-Hepatology and Nutrition

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Table 1
Participant characteristics.

	Celiac group	Control group	*p*
Number (*n*)	110	41	
Gender (girl/boy)	69/41	22/19	0.56
Age (months)	135.6 ± 37.0	124.2 ± 32.6	0.07
Height (cm)	138.9 ± 16.4	138.3 ± 13.8	0.80
Weight (kg)	35.3 ± 12.5	34.0 ± 13.1	0.57
Duration of disease (months)	39.2 ± 25.6	–	

with biopsy-diagnosed CD and 41 age and sex-matched controls (82 ears) were included in the study. Detailed histories were taken from participants in both groups to rule out secondary causes of hearing loss, and informed consent was obtained for all participants before the study. This study was performed in accordance with the principles of the Helsinki Declaration as revised in 2008. Exclusion criteria were history of intrauterine infection, perinatal hypoxia, ototoxic drugs, head trauma, ear surgery, diabetes mellitus, and a family history of conditions that can cause hearing loss. Age, sex, height, weight, and duration of disease were recorded for all CD patients, and CD was diagnosed according to the criteria of the European Society of Pediatric Gastroenterology, Hepatology and Nutrition with biopsy specimens analyzed according to the Marsh–Oberhuber classification [14].

Participants in both groups underwent otoscopic examinations, and those with normal findings were included in the study. All audiologic tests were performed by a single audiometrist. Tympanometry (GSI 33 Middle Ear Analyzer) was performed in all participants. Those with normal type A findings on tympanometry were included in the study.

Air conduction thresholds at 250, 500, 1000, 2000, 4000, and 8000 Hz and bone conduction thresholds at 500, 1000, 2000, and 4000 Hz were measured by pure tone audiometry (Interacoustics AC 40 Clinical Audiometer, Denmark). The pure tone average was calculated by taking the averages of thresholds detected at 500, 1000, and 2000 Hz. Hearing loss was considered above 20 dB and was defined as mild at 21–30 dB, moderate at 31–60 dB, severe at 61–90 dB, and profound at >90 dB. Participants with conductive hearing loss were excluded from the analysis.

2.1. Statistical analysis

The Kolmogorov–Smirnov test was used for checking normal distribution of continuous variables. The Mann–Whitney *U* test was used for two independent groups with non-normally distributed variables. Frequency, percentage, and mean ± standard deviation (SD) values were used as descriptive statistics. SPSS for Windows version 22.0 software was used for statistical analyses and $p < 0.05$ indicated statistical significance.

3. Results

The mean age was 135.6 ± 37.0 months in the CD group and 124.2 ± 32.6 months in the control group. There were 69 girls (62.7%) and 41 boys (37.3%) in the CD group and 22 girls (53.7%) and 19 boys (46.3%) in the control group. There were no significant differences between groups in terms of age, height, weight, and gender distribution ($p > 0.05$) (Table 1).

No air-bone gap was detected in any of the participants in either group.

There were no significant differences between the right and left ear thresholds in each category ($p > 0.05$).

There was a significant difference between the CD patients ($n = 110$) and the controls ($n = 41$) in right ear pure tone bone conduction thresholds at 500 Hz (B500) ($p < 0.05$). However, the difference was not sufficient to define clinical hearing loss, as the pure tone average was below 20 dB.

There were also significant differences between the CD patients ($n = 110$) and the controls ($n = 41$) in left ear pure tone bone conduction thresholds at all frequencies ($p < 0.05$), but no hearing loss was indicated by the pure tone averages for either group. There was a significant difference in the pure tone averages for bone conduction between groups, ($p < 0.05$), but no significant difference in the pure tone averages for air conduction (Table 2).

When the CD patients were divided into two groups according to duration of disease (≤36 months and >36 months), significant differences in bone conduction thresholds were detected at all frequencies ($p < 0.05$) (Table 3), with significant increases at the later stages of CD. However, this difference was not sufficient to define clinical hearing loss as the pure tone average was below 20 dB.

4. Discussion

The pathogenesis of neurological damage in patients with CD has not been fully understood, and the role of the gluten-free diet remains controversial [15]. It has been suggested that humoral immune mechanisms contribute to the pathogenesis of ataxia and peripheral neuropathy [16].

Nutritional deficiencies due to malabsorption have also been proposed as factors in the development of neurological deficits in patients with untreated CD. However, because improvement of

Table 2
Pure tone audiometric findings.

Hearing frequency	Celiac group (*n* = 220 ears) Mean ± SD			Control group (*n* = 82 ears) Mean ± SD			*p*
	Right ear (*n* = 110)	Left ear (*n* = 110)	Total ears (*n* = 220)	Right ear (*n* = 41)	Left ear (*n* = 41)	Total ears (*n* = 82)	
Bone500	13.1 ± 4.6	13.5 ± 5.6	13.3 ± 5.1	11.3 ± 4.3	11.1 ± 4.3	11.4 ± 4.6	0.003
Bone1000	12.5 ± 4.7	13.4 ± 5.5	12.9 ± 5.1	11.5 ± 4.4	11.2 ± 4.4	11.5 ± 4.6	0.024
Bone2000	12.7 ± 4.7	13.4 ± 5.4	13.0 ± 5.0	11.3 ± 4.5	10.9 ± 4.2	11.2 ± 4.5	0.005
Bone4000	12.6 ± 4.6	13.5 ± 5.5	13.0 ± 5.1	11.3 ± 4.3	10.6 ± 4.4	10.9 ± 4.3	0.001
Air250	17.2 ± 2.7	17.5 ± 3.0	17.3 ± 2.8	17.8 ± 4.8	19.0 ± 4.8	18.4 ± 4.7	0.064
Air500	16.6 ± 2.9	16.8 ± 3.2	16.7 ± 3.0	16.5 ± 3.9	17.2 ± 3.9	16.8 ± 3.8	0.793
Air1000	14.9 ± 4.4	14.6 ± 4.6	14.7 ± 4.4	15.1 ± 4.7	14.5 ± 4.4	14.8 ± 4.5	0.878
Air2000	14.3 ± 4.6	14.3 ± 4.6	14.3 ± 4.6	14.3 ± 4.7	13.8 ± 4.8	14.0 ± 4.7	0.657
Air4000	14.7 ± 4.7	15.0 ± 4.5	14.8 ± 4.5	14.6 ± 5.4	14.3 ± 4.7	14.4 ± 5.0	0.541
Air8000	15.4 ± 4.9	16.1 ± 5.5	15.7 ± 5.2	16.0 ± 6.4	15.6 ± 4.8	15.7 ± 5.6	0.953
PTA* for bone	12.8 ± 4.6	13.4 ± 5.5	13.1 ± 5.0	11.4 ± 4.3	11.6 ± 5.0	11.4 ± 4.6	0.010
PTA for air	15.3 ± 3.6	15.3 ± 3.8	15.3 ± 3.7	15.3 ± 4.0	15.2 ± 3.8	15.2 ± 3.8	0.865

PTA*: pure tone average.