

Chapter 20

Acceptability Analysis, Cultural Aspects and Personal Impact of Diagnosis

Eduardo Cueto Rúa¹, Luciana Guzmán¹, Cecilia Zubiri¹, Gabriela Inés Nanfито¹, María Inés Urrutia², Leopoldo Mancinelli³

¹Pediatric Gastroenterology. Division of Gastroenterology, Hospital Sor María Ludovica, La Plata, Argentina.

²Scientific Calculist, Gastroenterology Division, Hospital Sor María Ludovica, La Plata, Argentina.

³Clinical Psychology Gastroenterology Division, Hospital Sor María Ludovica, La Plata, Argentina.

cuetorua.eduardo@gmail.com, guzman_155@hotmail.com,
cecizubiri03@hotmail.com, gnanfито@hotmail.com, urrutia@isis.unlp.edu.ar,
leopoldomancinelli@gmail.com

Doi: <http://dx.doi.org/10.3926/oms.225>

How to cite this chapter

Cueto-Rúa E, Guzmán L, Zubiri C, Nanfито GI, Urrutia MI, Mancinelli L. *Acceptability Analysis, Cultural Aspects and Personal Impact of Diagnosis*. In Rodrigo L and Peña AS, editors. *Celiac Disease and Non-Celiac Gluten Sensitivity*. Barcelona, Spain: OmniaScience; 2014. p. 407-434.

Abstract

The authors have performed 1500 surveys on the subject of *acceptability of the celiac condition*, based on a questionnaire on the frequency of various claims by the celiac community. The questionnaire design resulted from previous meetings in which the authors participated as guest experts. The authors intended to characterize the subjective reactions of the attendants to said meetings. With that in mind, an investigation was made on the age, gender, educational level, number of celiac relatives, time and degree of adherence to the gluten-free diet, clinical features at the beginning of treatment, what issues patients find to be troublesome in every-day life, their wishes when confronted with this peculiar condition, their fantasies and their realistic expectations.

It was found that most of the participants were female and that educational level has a positive impact regarding a complete adherence to the diet. The number of persons with CD in the same family conspires against diet adherence and the most important social issue was not having restaurants with a gluten-free menu.

It was also discovered that the attendants' hoped-for solution was a cure and that their realistic expectation was to have safe, palatable and affordable food throughout the whole country as well as a law that took notice of their condition. When the 1306 surveys on the *Impact of Diagnosis* were analyzed, it was found that the exact words used by the physician and the patient's level of education do not play a key role in the time it takes to accept the celiac condition. In addition, it became clear that the words used by the physician have different effects when first heard by an adult with celiac disease and when first heard by a mother of children with celiac disease. These words, according to the authors' analyses, contribute to heighten emotions in patient support groups but definitely hinder the acceptance process.

1. Introduction

As pediatricians, the authors have always held that the concept of celiac disease is composed of words and gestures. For this reason, the physicians' attitude, at the time of diagnosing a patient and during the rest of the clinical management, must be carefully restrained. The purpose of this is to have a positive impact in the perception of this ailment, which will impose a lifestyle on young patients.

A sense of caring towards the patient must be taught to the patient's parents and family. If a two year-old patient is constantly addressed using words that denote compassion or pain and if it is suggested, through expressions and gestures, that these feelings are harbored for him or her, it will be hardly possible to expect this person to feel fully confident during his or her future development. Unknowingly, the authors have acted according to Pedro Lain Entralgo's (1908-2001) postulates which state that "A physician must be able to walk in his patient's shoes in order to feel what his patient feels and finally to help the patient overcome his difficulties".

The fact of thus conceiving the problem and the implications of intolerance to a foodstuff which, in Western culture, has symbolic and even (occasionally) religious associations led the authors to found, in 1978, the *Argentinian Celiac Disease Association*, the first one in America and the second one world-wide.¹

Inspired by the teachings of Dr. Horacio Toccalino (1931-1977) and having more than 40 years of history, the authors' group has investigated celiac disease and many of its different aspects, among which it is possible to highlight the establishment of a precise and objective mathematical relation to the degree of enteropathy according to the villus-crypt ratio, which has been efficiently used for over 30 years without modifications²; the discovery, in 1985, of the auto anti-smooth muscle antibody values³, of their variability in the challenge test⁴ and also (according to diet compliance) using them since that date in screening asymptomatic family members.⁵ The authors also underline the fact that they have employed a clinical and laboratory scoring system which has allowed to efficiently estimate the probability of suffering from celiac disease.^{6,7} In addition, the Argentinian Ministry of Health has proposed that it be used⁸ and that it be available online at no cost. Lastly, the social situation of the celiac disease patient has been taken into account. The authors have helped elaborate the current laws regarding the subject and have deeply studied and analyzed the patients' subjective reactions, needs and demands.⁹

Due to the above, the authors were invited to contribute this chapter.

Between March 2008 and December 2009, 1500 surveys of celiac disease patients were performed among those who attended the meetings of the Argentine Celiac Disease Association, trying to assess a number of social and cultural aspects. Between August 2011 and December 2012, another survey to evaluate the impact of diagnosis was performed.

It must be stated that Argentina has a high Celiac Disease (CD) prevalence, quite similar to that found in European countries. This country, especially the large cities along the Atlantic Coast and Río de la Plata and its tributary streams, has been the host of massive immigrations from Europe, especially from Spain and Italy. Additionally, Argentina is a great wheat producer, exporter and

consumer, baseline conditions favorable for the development of the disease. Regarding CD prevalence, a study by Dr. JC Gómez et al., carried out in couples that underwent the prenatal testing between 1999 and 2000, revealed a CD ratio of 1/167.¹⁰ On the other hand, a multicentric test performed by Dr. M. Mora and collaborators on children who were subjected to pre-surgical tests for scheduled surgery, or from emergency cases or else for physical aptitude tests, revealed a prevalence of 1/79.¹¹ In the Gastroenterology Service at the Interzone Hospital, specialized in Pediatrics in the city of La Plata, after reviewing the data base, the authors found that, from January 1st, 2000 to December 31st, 2010, 852 patients with a male/female ratio of 2/1 had been diagnosed.

2. Acceptability

In the first survey, named *acceptability*, the following subjects are discussed:

- 1) Age.
- 2) Gender.
- 3) Number of celiac disease patients at home.
- 4) Order of birth of the celiac patient (firstborn, middle child, last child or only child). The authors added one more parameter: whether a child is born separated by six or more years from the preceding sibling, who in turn is classified as last-only child.
- 5) Number of years of compliance with the gluten-free diet.
- 6) Clinical presentation at the time of diagnosis.
- 7) The patient's educational level.
- 8) The patient's parents' educational level, whether the former is a child or an adult.
- 9) Dietary transgressions.
- 10) Frequency of transgressions.
- 11) Location where said transgressions occur.
- 12) Whether they happen while alone or with company.
- 13) What kind of unsuitable food the patient desires to consume.
- 14) What product prompted the transgression.

- 15) Other incitements.
- 16) Whether the patient seeks help from support groups.
- 17) Whether the patient finds support or if he feels ill-treated in support groups or by his family.
- 18) What the patient dislikes about CD.
- 19) The patient's fantasies regarding the disease.
- 20) The patient's expectations regarding this clinical entity.
- 21) The patient's province of residence.

In order to achieve its goal, the survey was carried out at the meetings of the Argentinian Celiac Disease Association and in its different branches throughout the country as well as by means of other NGO's that support celiac disease patients and their families.

This project was undertaken in eleven Argentinian provinces and yielded the following distribution:

- Province of Buenos Aires: 57.8%
- City of Buenos Aires: 13.1%
- Santa Fe: 7.6%
- San Luis: 4.9%
- Córdoba: 4%
- Tierra del Fuego: 3.3%
- Neuquén: 2.9%
- Salta: 1.8%
- Other provinces: 4.3%.

It is noteworthy that when the authors, as health care professionals, participated in the meetings, the attendance was twice or even thrice than what would have been expected without their attendance.

To assess diet compliance after the surveys were done, the authors established a score ranging from 1 to 10. A score of 10 was given to those patients who rigorously complied with diet consuming only suitable products. A score of 7 was given to those who transgressed the diet about once a year. A score of 3 was obtained by those who committed transgressions on a monthly basis. A score of 2 was given to those who committed transgressions weekly and finally, a score of 1 was given to those who consumed gluten daily. This allowed to grade the level of diet compliance in each province. The ones analyzed were those in which more than 25 people were surveyed.

The results were as follows:

- Córdoba, with 60 participants who scored an average of 8.71%.
- Santa Fe: 114/8.62%.
- City of Buenos Aires: 196/8.6%.
- Province of Buenos Aires: 866/8.08%.
- Tierra del Fuego: 50/8.04%.
- Salta: 27/7.81%.
- Neuquén: 43/6.04%.

Age groups were used to classify the people surveyed:

- Earliest infancy (1 to 5 years old).
- Second infancy (6 to 12 years old).
- Adolescents (13 to 17 years old).
- Young adults (18 to 40 years old).
- Adults (41 to 60 years old).
- Older adults (over 61 years old).

This afforded a singular understanding of the celiac disease situation.

So far, it is clear that Celiac Disease is a permanent gluten intolerance, but it is necessary to remember that the protein itself comes from a foodstuff with an enormous cultural symbolism and that it is legally added to a vast number of industrialized products. This toxic protein, which generates celiac disease in the patient, is found in wheat, oats, barley and rye, a fact that became known through the scientific research of Dicke, Weijers and Van de Kamer^{12,13} (worthy of a posthumous nobel prize).

The main issues to be addressed were: What happens during and after the moment when the physician diagnoses a patient? What impact does it have on the patient's life? How does this affect the patient-physician relation? Is the diet properly adhered to? If any, which are the reasons for giving up and transgressing the diet? The authors were also interested in determining whether the patient's educational level (and that of the patient's parents) somehow influenced diet compliance.

The concept of Celiac Disease is something that has to be constructed, and the first brick of this structure is placed by the physician at the time of the diagnosis.

The words used, the facial expression, the poise shown, the fact of having arrived to a positive and final diagnosis with implication of having solved one problem and not witnessing the beginning of another are indispensable for the patient to feel that this is the start of a better life and not the starting point of a series of adversities and sufferings.

Once the patient has been diagnosed, it is important thing to control his or her food, which must be based on a diet without wheat, oats, barley and rye. A physician takes 7 seconds to prescribe

a diet that the celiac disease patient will have to follow for the next 70 years. Out of the thousands of foodstuffs humans have at their disposal, only four are prohibited (wheat, oat, barley and rye). One of them, barley, is currently being studied, with conflicting results.^{14,15} As Real and collaborators demonstrated, “these results suggest that the oats have a wide variation range of their immunotoxic potential, which could be due to the differences in the immunogenicity degree in its sequences”.¹⁶

The wide usage of gluten-bearing cereals in the West makes diet compliance challenging. This diet, which seems theoretically simple to follow, becomes, in practice, a trial for the patients who must follow it¹⁷ and for the physicians and dieticians who must collaborate with and guide the patient.

Gluten consumption, for the celiac disease patient, voluntary or not, carries the potential risk of associated diseases, which is why the patient must be warned of this risk.¹⁸

After having thoroughly analyzed the survey variables, some observations come to mind. The global acceptability percentage for the Gluten Free Diet (GFD) among people who attend to support group meetings hovers around 70%, probably being for this reason slightly higher than in the reviewed bibliography¹⁹; however there are countries with a strong celiac disease culture in which diet compliance is stronger.²⁰

The adolescent population represents a challenge. Many publications echo this view.^{21,22} Adolescence constitutes a period in which the gluten-free diet is frequently abandoned. For this reason, regular medical examination is imperative, as are serum antibody determination and testing for signs and symptoms that can reveal nutritional deficiencies and other autoimmune diseases.

Regarding the situation of celiac disease patients living in the same house, it would seem that when only one family member is afflicted, GFD compliance is very effective and it would seem that the family actually plays a protective role. When several family members suffer from celiac disease, the transgression of one can trigger a domino effect in the rest of the affected family members. It is here where the greatest failure rate is seen: There seems to be a permissive attitude among family members. Even if some patients, after transgressing the diet, show symptoms like headaches, vomiting or abdominal pain, it is also true that there are many who remain asymptomatic. It should be pointed out that in those families with more than one celiac disease patient, these last were identified through family screening, which is the reason why these patients have never felt ill and the reason why their transgressions did not have any clinical manifestations.

When analyzing how the birth order is related to compliance with the GFD, it is reasonable to conclude that single children are better in adhering to the treatment. These children have a family that focuses exclusively on them. In the case of last-only children, those whose preceding sibling is at least six years older, families have a different behavior since they might have been the outcome of an unexpected pregnancy, a fact that has strong connotations for the mother. Generally, these children are pampered, probably because of the sense of responsibility felt by the parents. It is not by chance that, the last-only child, is worse in adhering to the diet, has more

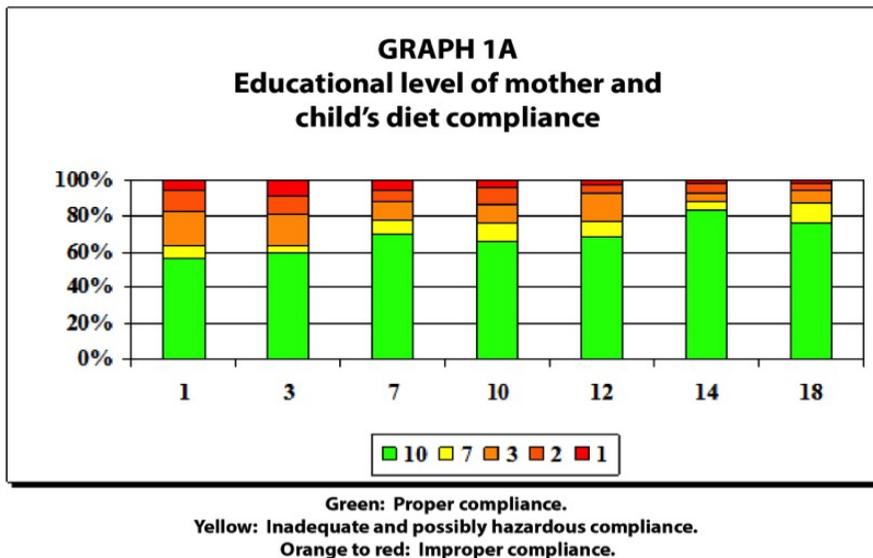
cavities (excessive consumption of sweets) is more often constipated (low fiber consumption and high dairy product consumption).²³⁻²⁴

This survey showed that earlier diagnoses correlate with a higher transgression percentage, as other authors have observed.²⁵

This would seem to be related to the lack of clinical expression of these transgressions and, therefore, since the patient does not feel ill, he or she continues to indulge in them.^{26,27}

As for the CD presentation form, it has been observed that it determines the degree of gluten-free diet compliance, but, contrary to expectations, those who exhibited the most severe initial symptoms do not adhere to the diet as thoroughly as those who have monosymptomatic diseases or those who were diagnosed through associated diseases. Furthermore, those who have lower adherence to the diet are those who are found through family screening. These patients, generally asymptomatic, do not perceive the disease and are unable see any benefit in following the diet.

In terms of the relation between educational level and the GFD, it seemed reasonable to conclude that patients with a higher educational level have greater access to information and to health services and, therefore, also possess the means to indulge in an expensive diet.²⁸ Another significant, noteworthy fact was that children or adults whose mothers had dropped out of university reached the highest percentage in diet compliance. Nevertheless, this peculiarity lacked a significant statistic value. It is not being suggested here that these studies be abandoned but, rather, that further care ought to be exercised in a similar situation (Graph 1A).



Graph 1A. Level of diet compliance and educational levels reached by the child's mother. When the first groups are compared (illiterate or incomplete primary school) with the last two groups (incomplete university or graduate) there is a significant difference. $p = 0.00001$.

Even though there is still a lot of controverted data to analyze, during the last decade a significant number of published articles have shown weighty evidence on the long-term efficacy of the GFD to prevent complications and undesirable associations.²⁹

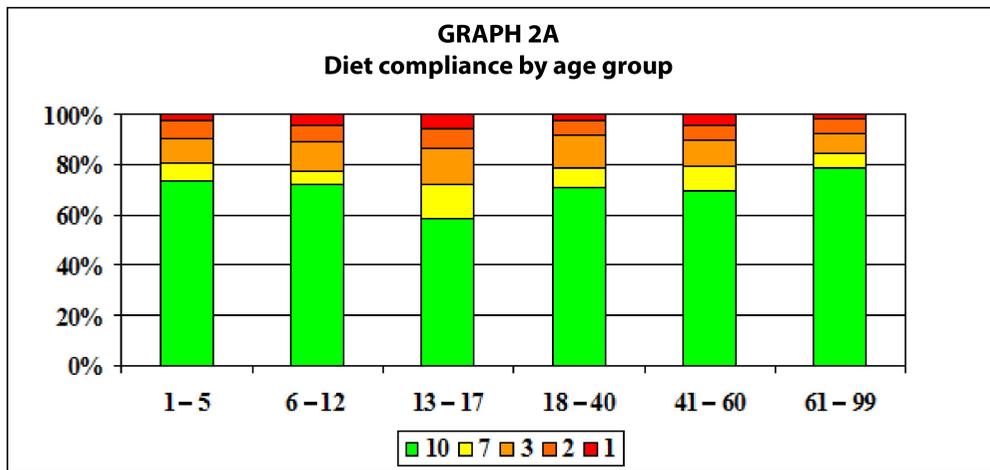
However, despite the benefits of the GFD, a percentage of patients decides not to adhere to it or else they do, but not strictly.³⁰ Ideally, celiac disease patients should be monitored regularly by a team that includes physicians, dietitians and psychologists. Nonetheless, the strongest support comes from celiac patients' associations who can empathize with the problem since they already experience it first-hand in their daily lives.

Finally, these surveys revealed that the celiac disease patient's longed-for solution is a cure for the disease through some scientific discovery; realistically, celiac disease patients desire safe and affordable foodstuffs available throughout the whole country and a law that truly takes into account the social problems generated by celiac disease.

2.1. Conclusions about Acceptability

Briefly, the conclusions of the acceptability study were as follows:

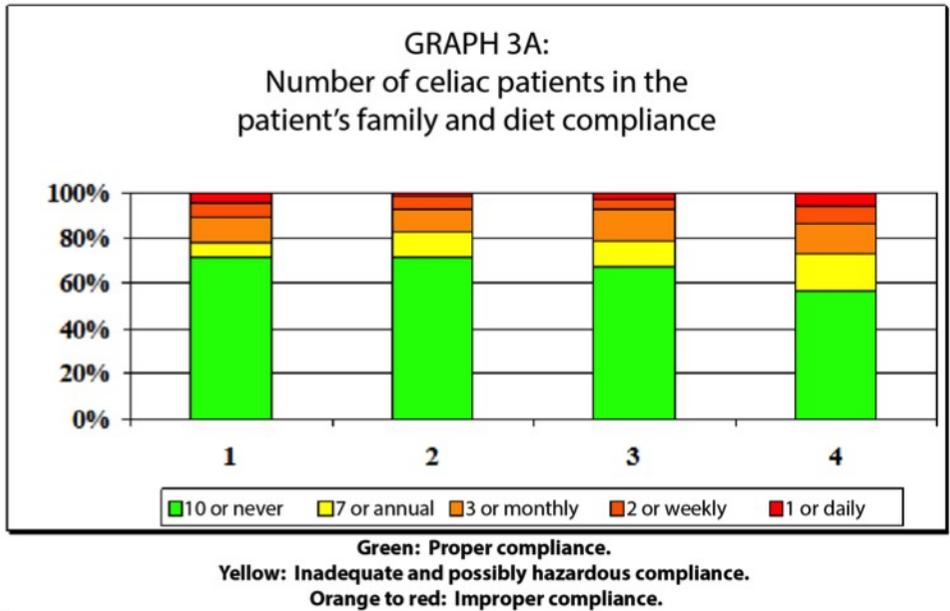
- a) GFD compliance was of 70%.
- b) GFD compliance is the same for each gender, whether in children or in adults.
- c) The worst score was seen in adolescents (Graph 2A).



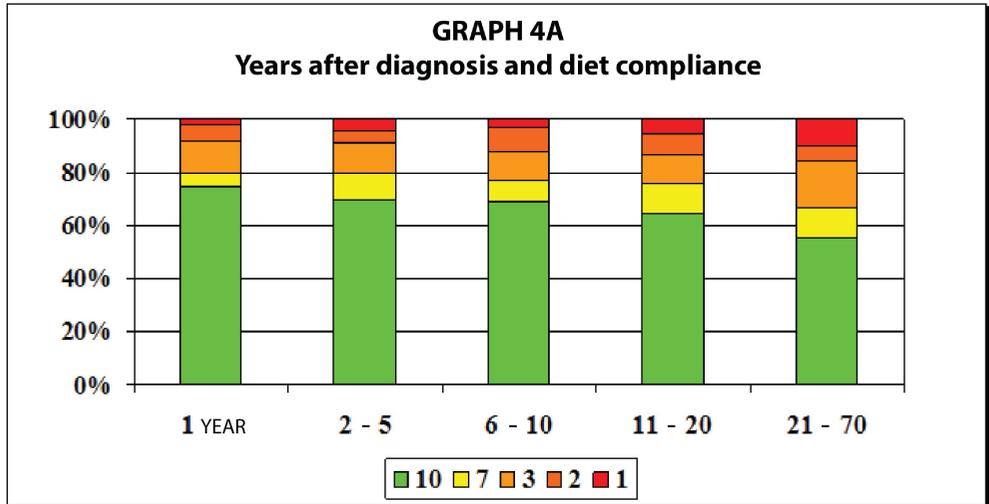
Green: Proper compliance.
Yellow: Inadequate and possibly hazardous compliance.
Orange to red: Improper compliance.

Graph 2A. Diet compliance level by age group. Highest diet compliance level is achieved by the 61 year-old or more group. Second is the group of 1-5 year-old children under the care of their mothers; the group with the worst compliance were the adolescents (13 to 17 years old).

- d) GFD compliance diminished as the number of family members with CD increased (Graph 3A).
- e) GFD compliance deteriorated as years elapsed from the time of diagnosis (Graph 4A).

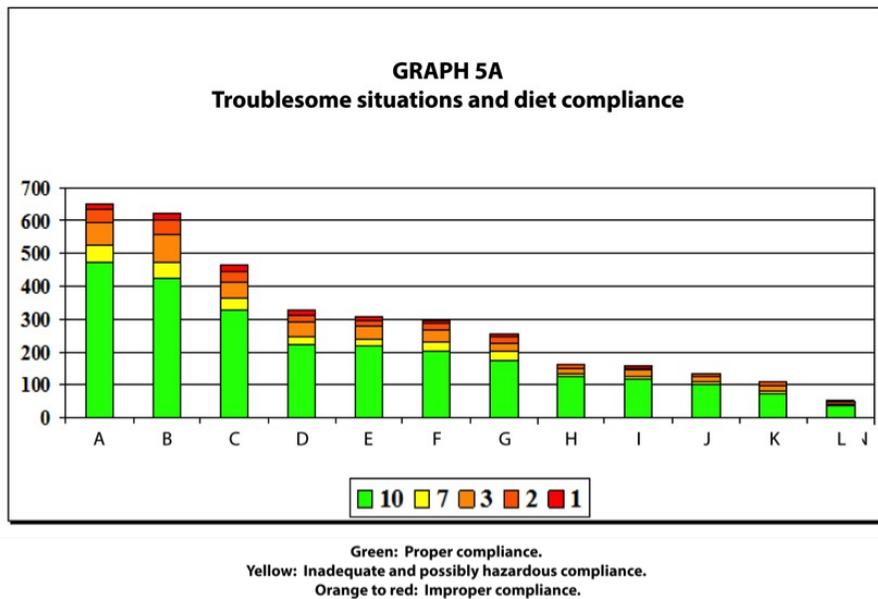


Graph 3A. GFD compliance level revealing a decline according to the number of members in the family. Statistically, this observation was significant. $p=0.04$.



Graph 4A. Gradual lack of diet compliance as years elapse. Increasing amounts of years are grouped; analysis of this phenomenon yields a highly significant value of $p=0.001$.

- f) In general, Gluten-Free Diet compliance level is better when educational level is higher. Nevertheless, this did not yield a significant statistical value.
- g) Best GFD compliance level was seen in single children, while the worst case was seen in last-single children.
- h) Highest treatment adherence was seen in the cases where the patient was diagnosed by finding monosymptomatic forms of celiac disease, the worst treatment adherence occurred in patients diagnosed through family screening.
- i) Most transgressions were committed at home.
- j) Products like bread, pastry and sandwiches were highly desired.
- k) Easily accessible and smaller products like candies and cookies proved to be an irresistible temptation.
- l) The four issues that are most troublesome for the patient are: (Graph 5A).



Graph 5A. A decreasing scale of the most bothersome situations for a celiac disease patient: A: Restaurants without a GFD menu; B: Being forced to carry their own food; C: Not finding gluten-free products in 24-hr stores; D: Being asked inappropriate CD-related questions; E: Not being taken into account by the private health system; F: Not receiving social benefits for their condition; G: Being made to feel different; H: Feeling a burden to the family; I: The lack of seriousness given to the subject by teachers; J: When a professional states that small amounts of gluten will not do any harm; K: When a professional states that there may be a cure; L: Patients has no complaints.

- 1) *Restaurants without a GFD menu.*
 - 2) *Having to carry their own food.*
 - 3) *Not finding gluten-free products in 24hr. stores.*
 - 4) *Being asked inappropriate CD-related questions.*
- m) The celiac disease patient's fantasy: a definitive cure.
- n) A desired, plausible reality: a law that fully contemplates the Celiac Disease condition.
- o) In general terms, diet compliance is satisfactory.

3. Second Survey: Sociocultural Aspects. Impact of Diagnosis; Terms Used by the Physician; the Patient's Emotions; the Patient's Educational Level and the Time Needed to Accept the Celiac Disease Condition

3.1. Introduction

Human relationships are diverse in nature; some are superficial, others are profound and some are of a singular complexity, such as the one between physician and patient. As Dr. Moreno Rodríguez stated, "the physician-patient relationship has been, is and will always be the most sensitive and human aspect of medicine".³¹ As pediatricians, making lifelong diagnoses has led the authors to consider the appropriate way to do this, to measure and analyze the words used, to try empathize with the patient and to reinforce what is said with the appropriate gestures and attitude, exhibiting a considerate but firm attitude. When the patient is advised that he or she must strive to make things happen because they won't happen by themselves, the moment when the diagnosis is made must also be considered.

The precepts for medical practice are found in the Hippocratic Oath, "...I will direct the diet with a view to the patients' recovery, according to my strength and judgment...", "...I direct my patients' recovery according to my strength and judgement".³²

The physician's characteristics in this relationship were summarized by Hippocrates more than 2000 years ago when he stated that the former must possess four main qualities: knowledge, wisdom, humanity and integrity. These qualities are well reflected in the three parameters established by Pedro Laín Entralgo: 1) To empathize with the patient, 2) To feel what the patient feels, 3) To be willing to help the patient when he or she faces difficulties.³³

In order to achieve these parameters, the physician must thoroughly know the etiopathogeny of celiac disease, its different presentation forms, how to reach diagnosis, its treatment and its complications; but the patients' feelings can only be known by listening to them and allowing them to express themselves. This is why the authors have undertaken different surveys and, from

these premises, dedicated themselves to the study of the impact of the diagnosis and to evaluate the physician's responsibility in it.

4. Analysis of diagnostic impact

In order to continue with the diagnostic impact analysis, a second stage was undertaken surveying 1306 people in support groups' meetings recognized in Argentina or at their branches.

To this end, three identical surveys were designed for: 1) Adults with celiac disease, 2) The patient's relatives and 3) Children with celiac disease who could answer it by themselves or with their parents' help.

The survey was anonymous and it included:

1. Age.
2. Gender.
3. Number of celiac disease patients in the house.
4. Years of following the diet.
5. Patient's educational level or his or her mother's:
 - a. 1 to 7: grade school
 - b. 8 to 12: high school
 - c. 14: incomplete university
 - d. 18: university graduate
6. Reaction to diagnosis, with the following possibilities:
 - 6.1. Happiness.
 - 6.2. Tranquility.
 - 6.3. Resignation.
 - 6.4. Upset.
 - 6.5. Sadness.
 - 6.6. Annoyance.
 - 6.7. Anguish.
 - 6.8. Anger.
 - 6.9. Fear.
7. How long did it take for the patient to accept the celiac disease condition?

8. Diet transgressions:
 - a. No transgressions.
 - b. Once a year.
 - c. Monthly.
 - d. Weekly.
 - e. Daily.

9. How does the patient feel in the support group
 - 9.1. Very good.
 - 9.2. Good.
 - 9.3. Neutral.
 - 9.4. Used.
 - 9.5. Hurt.
 - 9.6. Out of place.

10. Where does the patient want to find or buy gluten-free products?
 - 10.1. Neighborhood stores.
 - 10.2. Supermarkets.
 - 10.3. Health-food stores.
 - 10.4. Pharmacies or drug stores.

11. City where the patient lives.

12. Province where the patient lives.

13. Words used by the physician when diagnosing (does the patient remember any of the following?):
 - a. A severe disease/misfortune.
 - b. A true disease.
 - c. Celiac Disease.
 - d. Food intolerance.
 - e. A diet solves the problem.

The surveyed patients' ages ranged from 1 to 84 years old. In the cases of those patients who were unable to answer, their parents provided the data. Percentages by province surveyed:

- | | |
|--------------------------------|------------------------------|
| a. Buenos Aires: 44.1% | g. Santa Cruz: 3.1% |
| b. Córdoba: 12.3% | h. Río Negro: 1.6% |
| c. City of Buenos Aires: 11.2% | i. Jujuy: 1.5% |
| d. Chubut: 10.9% | j. San Luis: 1.3% |
| e. Entre Ríos: 7.0% | k. La Pampa: 1.2% |
| f. Corrientes: 3.1% | l. All other provinces: 2.6% |

People from 164 different cities participated in this survey.

- 1) Age, gender and number of celiac disease patients in the family were analyzed, making distinctions between adults, patients' relatives and children with celiac disease (Table 1).
- 2) Patients were grouped according to the number of years of adherence to the diet and their distribution was analyzed percentually, which yielded six groups, among which those who participated the most were those patients who have been following the diet for 2 to 5 years (Table 2).

	Celiac adults 58.6%	Celiac relatives 36.4%	Celiac children 5%
Number 1306	765	476	65
Gender	F 79.2% M 18.8%	F 67.4% M 32.6%	F 68.8% M 31.2%
Mean age	42.5 years		11.3 years
DS	+/- 14.04		+/- 3.00
Educational level			
Mean	13.4		7.2
DS	+/- 3.64		+/- 3.13
Median	14		8
Mode	12		8

F: female, M: male

Table 1. General data on the surveyed population. The celiac patients' relatives category is variegated since its members included both genders and all age groups, thus a mean cannot be found for neither their ages nor their educational levels.

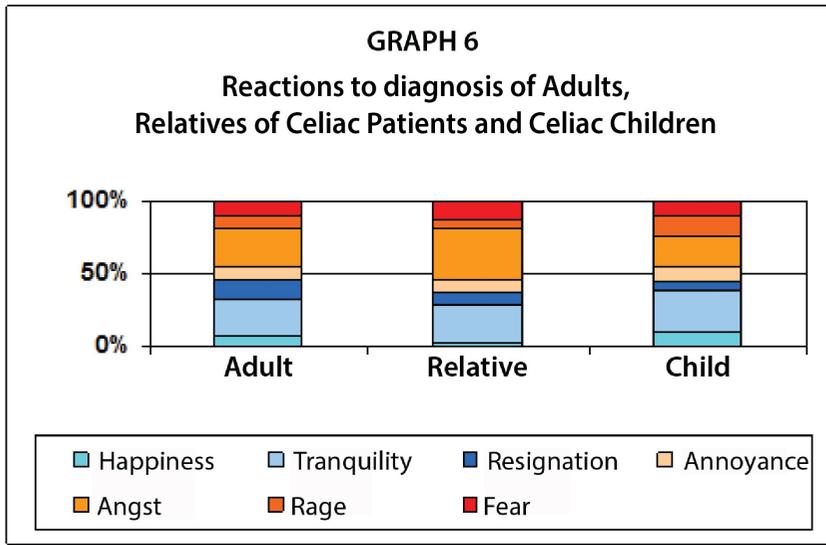
1306 cases	Celiac adults 48.6 %	Celiac relatives 36.4%	Celiac children 5%
0 years	13.7 %	11.9 %	12.5 %
1 year	17.4 %	19.9 %	20.3 %
2 a 5 years	37.9 %	39.0 %	40.6 %
6 a 10 years	17.9 %	20.3 %	20.3 %
11 a 20 years	8.3 %	8.1 %	6.3 %
21 years or more	4.7 %	0.8 %	0.0 %

Table 2. Distribution according to years of following the diet. The group with the highest meeting attendance is the group which has 2 to 5 years of adhering to the diet. If we add the following group, 6 to 10 years adhering to the diet, between them both account for more than 50% of the attendance.

3) As for reaction to diagnosis, there is a dependence between this variable between being an adult or a relative of a celiac child, with a significant difference ($p= 0.001$). It can be seen that emotions like anguish and tranquility have higher percentages. When exclusively analyzing these last two survey percentages, it becomes noteworthy that, when relatives of celiac patients answer (mostly mothers), the answer is anguish. In contrast, in adults and children, the larger percentage expresses tranquility (Table 3, Graph 6).

Reaction to diagnosis 1306 cases	Celiac adults 58.6%	Celiac relatives 36.4%	Celiac children 5%
1-Joy	7.9%	3.7%	9.8%
2-Tranquility	24.6%	24.6%	29.5%
3-Resignation	14.3%	8.9%	6.6%
6-Annoyed/upset	8.0%	9.3%	8.2%
7-Anguish/sadness	26.2%	34.6%	21.3%
8-Anger/fury	8.6%	5.4%	14.8%
9-Fear	10.4%	13.6%	9.8%

Table 3. Reactions to diagnosis. The prevailing emotions are anguish and tranquility in all three groups. In the children’s group, tranquility prevails; in the other two, anguish prevails.



Cool tones: Positive response.
Warm tones: Negative response.

Graph 6. Diagnosis impact. Although they have similar reactions, the patients’ relatives group is slightly ahead of the rest in terms of annoyance, anguish, anger and fear.

- 4) Regarding the time required to accept celiac disease, it can be seen that children are the fastest to accept their new condition. When analysis was made considering acceptance during the first six months, the percentage difference seen in adults was statistically significant ($p=0.023$).

Time it takes to accept CD 1306 cases	Celiac adults %	Celiac relatives %	Celiac children %
1 week	42.6	43.4	53.8
1 month	18.5	24.7	16.9
6 months	10.5	7.4	15.4
1 year	9.6	11.3	4.6
Several years	7.2	5.4	4.6
Never	11.6	7.8	4.6

Table 4. Time elapsed before celiac disease acceptance. Acceptance is of over 60% among all three groups in a span of a month or less.

However, in the first month, the children’s acceptance reached 70.7% while adults’ stayed at 61.1% (Table 4).

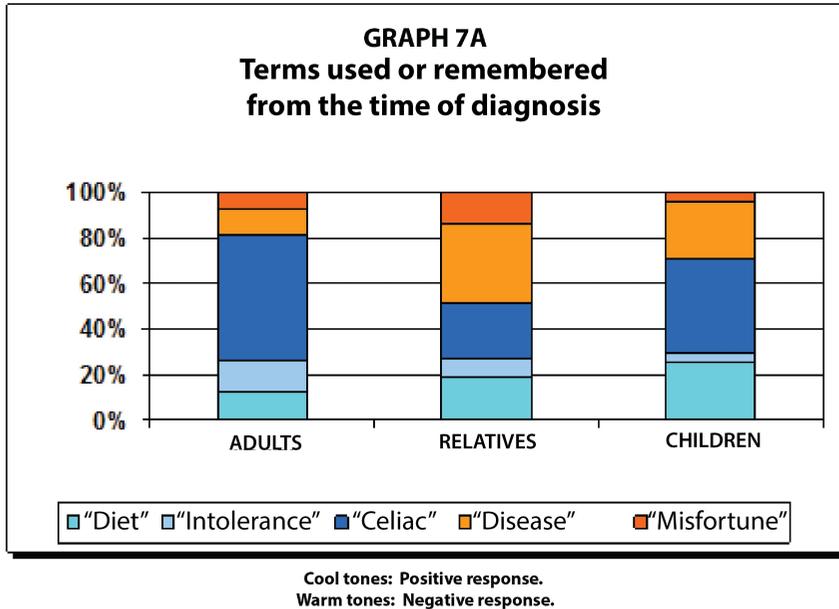
- 5) If we analyze the words expressed (or those the patient thinks he or she remembers at the time of diagnosis), “misfortune” is the one found to be most significant for relatives (mother). On the other hand, regarding the term “Celiac Disease”, mothers remember (or believe they remember) that the diagnosis was communicated or perceived as “disease”, while adults and children related it mostly to the word “celiac”.

Furthermore, comparing the mothers’ perception percentages to those of adults, the difference is highly significant ($p= 0.000$) (Table 5, Graph 7).

Terms used or recalled	Adults with CD %	Celiac relatives %	Celiac children %
“Misfortune”	6.9	13.9	4.2
“Disease”	11.5	34.3	25.0
“Celiac”	55.4	24.1	41.7
“Intolerance”	13.7	8.8	4.2
“Diet”	12.4	19.0	25.0

Table 5. Words used or remembered by the patient or patient’s relative when receiving the diagnosis.

Another noteworthy point was that the words used by the physician, or their interpretation by the patient, did not significantly influence the time it takes to accept the disease. It can be also said that the only advantage of term “misfortune” is that it has the highest compliance percentage but also the disadvantage of having the highest percentage of lack of acceptance of the disease.

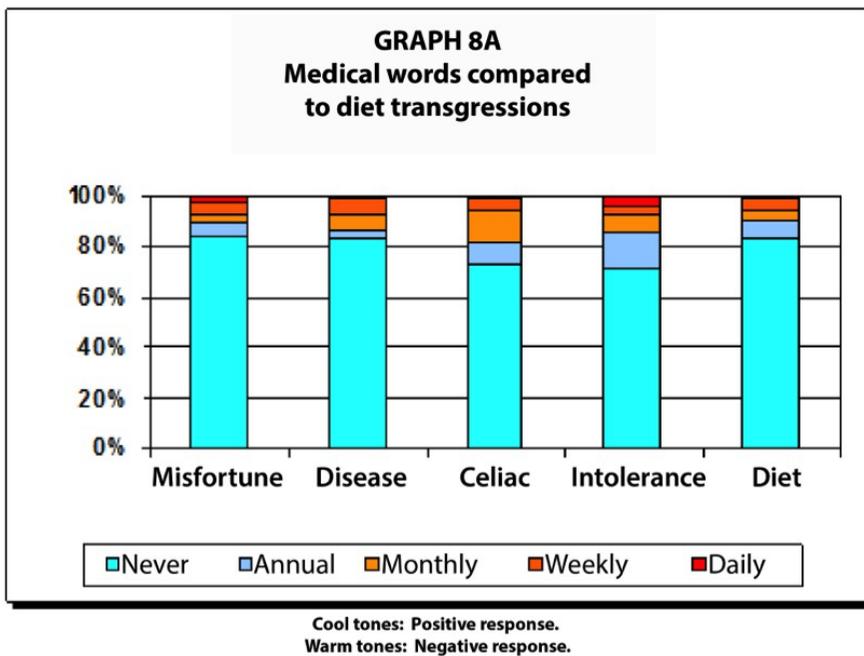


Graph 7. Impact. This graph shows that, among the words used by the physician, or as recalled by the mother or relatives, the word “disease” is the most important. Conversely, adults and children remember the word “celiac”. “Misfortune” has a greater impact on the patient's relatives.

- 6) Comparison between groups with elementary and higher education, regarding acceptance in a span of one week or not at all, was not significant. Acceptance time, in this survey, seems to be more related to personal history or patient's background than to educational level. The same thing happens when comparing the words remembered to have been used by the physician at the time of diagnosis, where educational level plays no role.
- 7) Generally speaking, the words used by physicians at the time of diagnosis and the level of diet compliance demonstrate that persons who thought to have a severe disease or remembered the diagnosis as a “misfortune”, actually adhered better to the diet than those with whom the term “intolerance” was used, with a highly significant high difference ($p=0.008$) (Table 6, Graph 8).

Words used / frequency of transgressions	Misfortune %	Celiac disease %	Celiac %	Intolerance %	Diet %
Never	84.2	83.6	73.0	70.7	83.0
Yearly	5.3	3.4	8.7	14.6	8.0
Monthly	3.5	6.0	12.8	7.3	4.0
Weekly	5.3	6.0	4.1	3.7	4.0
Daily	1.8	0.9	1.4	3.7	1.0

Table 6. Relation between the words uttered by the physician and different transgression levels.



Graph 8. Impact. The words “misfortune” and “disease” have a satisfactory impact on diet compliance, as opposed to the word “intolerance”, which exhibits correlation with poorer diet compliance.

- 8) Regarding the way patients felt at self-help group meetings, the well-being index rose to 92%.

9) Regarding the locations where celiac disease patients were most interested in acquiring their food, the results were:

Supermarkets/stores:	51.2 %	Health food stores/supermarkets:	41.6 %
Stores/health food stores:	3.8 %	Supermarkets/pharmacies:	2.4 %
Stores/pharmacies:	0.5 %	Health food stores/pharmacies:	0.5 %

Supermarkets alone had a total of 95.2%, local stores 55.5%, health food stores 45.9% and pharmacies only 3.4%.

Briefly, the conclusions of the impact survey are:

1. Reaction to diagnosis was different in relation to the group to which they belonged. Patients, adults or children with celiac disease react differently from the patient's relatives (mothers).
2. The emotion prevalent among adults with celiac disease and patients' relatives (mother) was "anguish", while in children with celiac disease the main one was "tranquility".
3. Even though most celiac disease patients rapidly accept their condition, children stood out significantly regarding their promptness to do so.
4. The terms employed by the physician at the time of diagnosis have a significant impact on the mother but not so in children.
5. The The terms employed by the physician also do not influence the time it takes to accept their condition.
6. The patient's educational level does not affect or modify the result of the variables analyzed here.
7. The urgency with which the physician states the diagnosis encourages the patient's relatives to have a better adherence to the diet and to have a positive experience in support groups. Exactly the opposite happens when the seriousness of the matter is depreciated, which correlates with the patients usually not feeling well in support groups.
8. The urgency with which the physician states the diagnosis does not have any impact on diet compliance among adult celiac disease patients.
9. The urgency with which the physician makes the diagnosis does not make adult patients accept their celiac disease condition in spite of adhering to the diet.
10. People mostly choose to buy their products at supermarkets; neighborhood stores are in a distant second place, followed by health food stores and, more distantly, drug stores.

It could be inferred from this last point that the celiac disease patient wants to live with his or her condition in a normal fashion and not associated with the health care system.

5. Celiac Disease: A Systemic Overview

Diagnosing a person with celiac disease generates a series of structural changes and readjustments within the family, which usually last a long time.

After receiving the news, nothing will be as it used to be within the family group: its functioning immediately abandons its habitual spontaneity and the simplest and most casual movements become burdened by a layer of complexity. What previously were simple outings and trips must now be the subject of a much more detailed attention and a series of preparations and often the plans must be aborted.

On the other hand, it is possible that the family group will divide itself into two clearly differentiated subsystems: one will be formed by the patient and the person who will provide him or her with logistic support (the mother if it is a child or an adolescent; the wife or husband if it is an adult). The other subsystem will be formed by the rest of the family.

The difference between these two subgroups obeys the change produced in interpersonal relationships since the moment the diagnosis is given, since the patient and his support have established a closer link, attend to medical consultation together, receive hygiene and diet advice and have begun to participate in support groups which discuss celiac disease.

Meanwhile, even if the rest of the family becomes acquainted with the new dietary and culinary rules, it could express lack enthusiasm regarding the treatment. So, it can be said, without fear of exaggeration, that the rest of the family may prefer to act unconcernedly rather than shouldering such a complex situation.

This difference between both subsystems, which does not necessary occur in all cases, but quite often, is triggered mainly by interpersonal due to the fact that not all family members empathize with the problem, even though it is clearly serious. These differences among family members generate discomfort.

5.1. Level of Adherence and Family Balance

Adherence to treatment depends in keeping an attitude of continuous alert to avoid eating food which cannot be tolerated by the celiac patient. In order to sustain this regimen through time, it is necessary that both subsystems start working as a team, involving the family as a whole in the treatment adherence. Thus, everyone should show interest in the details regarding the diet and culinary resources.

A family which adheres to the treatment would try by all means to encourage and keep adherence, and so cancelling the gap that initially separated both subsystems.

When this is not possible, the gap between both subsystems will only widen, conflicts will increase and the system tries to save itself by sacrificing its weaker link: treatment adherence. The diet is interrupted and the family prefers to ignore the situation.

5.2. Treatment Adherence and Adolescence

As it occurs with chronic pathologies and organ transplantations, adolescent celiac disease patients may exhibit attitudes of resistance, rebelliousness and diet neglect. This is caused by the hostile emotions generated by being compelled to follow a strict diet, barred from the enjoyment which others have with no limitations; the adolescent must also face the conflicts and discomforts of a stage in his or her life when he or she feels willing to confront and defy his or her elders. An obvious and infallible way to achieve this by not following the GFD.

The adults, who by now are getting used to the rigors of the treatment, now see themselves forced to face another source of conflict: the celiac patient's adolescence. In the ensuing tug-of-war between adults and adolescents, the latter can pressure the adults by threatening to abandon the treatment supported by allies external to the family, such as their group of friends or peers.

In spite of having assurance of their parents' affection, adolescents have a strong necessity to be part of a group and to earn its approval. Therefore, an adolescent will not hesitate to act in a way that pleases their peers, even when this may be disagreeable to his or her parents. The lack of treatment adherence in this phase can be furthered by a lack of parental skill.

5.3. Extended family and treatment adherence

The extended family can exert an important influence regarding the way treatment is faced, which will vary depending on the type of relationship previously established between the celiac patient's family and its relatives, as well as on the degree of proximity between them. It often happens that certain gaps in treatment adherence may have been provoked by messages from older adults who disbelieve the diagnosis and who consider the prohibition to consume bread, a foodstuff traditionally held to be beneficial and which has religious associations, to be illogical. Fortunately, situations like these are greatly decreasing in Argentina.

It must be taken into account that some suggestions offered by older, trustworthy relatives, which are repeated with regularity, can exert great influence, above all if they are compounded by the enjoyment implied by abandoning the diet. The advice or suggestions offered such relatives, which stems from their personal experience, can be well received by the younger relative, who may react favorably to an excuse to satisfy his or her oral desire.

Thus it is important to take into account the opinions of all the family members in order to avoid well-meant but counterproductive messages.

5.4. Treatment Adherence and Mass Media

An important adjuvant to treatment adherence comes from the TV programming which contain, culinary and kitchen hygiene suggestions and methods, as well as testimonials from celiac patients and stories which awaken the audience's interest, whether they tolerate gluten or not, and demonstrate a diet which will be palatable for all and which will cast aside the idea of a bland, elemental diet. Thus, the gap between tolerant and intolerant patients starts to narrow in such a way that the former may feel tempted to try the latter's diet.

Media coverage of the constant testimonies of a large number celiac patients, be they famous or anonymous, show us a full and pleasant life with total treatment adherence and allows an amelioration of the negative aspects of the celiac disease condition. It must be admitted that, in celiac disease, there is a series of systems that reciprocally influence each other and the most powerful of these, mass media, exerts a decisive influence on the pathology and in the way it is perceived. Through them, gluten intolerance is shown to mass audiences and given a widespread character, legitimacy and prestige.

5.5. Promoting adherence to treatment

Since adherence to the gluten-free diet is the treatment's cornerstone, efforts to that end start since the very first medical consultation, when diagnosis and treatment conditions are expounded and maintained. These injunctions are directed to the celiac patient but they must equally reach all the members of his or her family system since all of them must be bearers of the physician's message and therefore they need to collaborate in this continuous and prolonged effort to adhere to the physicians' prescriptions. It must be borne in mind that this intolerance is expressed against a ubiquitous substance which is found almost all edible products and which is not easily detectable, which is the reason why the patient, besides having a firm conviction regarding the diet, must be alert in all situations involving the consumption of food.

Adherence to treatment can sometimes falter due to periods of anxiety, frustration and anger triggered by painful circumstances or by psychological conditions reactive to stressing environmental factors. The patient appeals to food as a safety valve against stress. Under these circumstances, the family system, which by now is acquainted with this kind of reaction must rush for a medical consultation to prevent the patient's reaction from becoming a habitual pattern.

5.6. Gluten and Tobacco: Celiac Culture and other People's Consideration

Intolerance to gluten and tobacco smoke exhibits, from a systemic point of view, some coincidences. Until the middle of the last century, non-smokers who wanted to travel, eat, or rest in open smoke-free spaces, had to explicitly require it and then they were directed towards separate locations. Now, the situation is inverted. Smokers must be confined to separate areas if they desire to eat and smoke in a restaurant and totally abstain from it while using public transportation or while travelling by airplane.

Nowadays, the situation of celiac patients in public areas resembles to that of the non-smokers of past decades. When they arrive in cities where celiac disease is barely known, or even unknown, they must somehow think of a way to gather the ingredients of a reasonable meal. It is also probable that in restaurants which offer gluten-free dishes are not fully conscious of the special care needed when handling kitchen utensils and which may not have appropriate experience regarding this kind of food.

Nevertheless, the situation is changing and gluten-free dishes seem more appetizing. The social media is full of advertisements and offers and it is foreseeable that in a not too distant future celiac patients will be able to enjoy the same comforts as non-smokers do today.

5.7. Acceptance of the disease

When we talk about the acceptance of this disease, the patient supposedly has gone through an assimilation, a sudden and clarifying confirmation about his or her situation, which radically changes his or her attitude towards treatment. This may be an idealized view of what actually is a process composed of advances and retreats negotiated with relevant persons in their environment. For this reason, due to the lack of a definitive conviction, the patient must always be motivated and monitored so that he or she will be kept on the lookout regarding his or her adherence to the treatment's prescriptions.

There are many daily circumstances that place the celiac patient in a difficult situation, making him or her complain about these restrictions, above all those that generate exclusion and set him or her aside from his or her peers. When attending a social gathering carrying his or her own food, the patient may feel that this situation sets him or her aside and that it brands him or her with an unwanted distinction.

It can be argued, logically, that after several years of following a gluten-free diet, the patient achieves such identification with his complex dietary routine that his social interaction occurs smoothly. In spite of this, it is probable that some personal problem or a minimal logistic inconvenience may prevent him or her from having access to the food he or she needs at the appropriate moment, which may trigger an outburst of anguish or rage against his or her gluten-intolerant condition.

It must be admitted that true acceptance of the celiac disease condition will only occur when gluten-free products become as accessible, affordable and appetizing as other mass-

consumption products. The lack of these or difficulty in obtaining them hurts the patient's self-esteem and casts him or her in the position of a second-class citizen. As the great national poem of Argentina wisely states: "The heart of he who must beg bleeds copiously" (Martín Fierro).³⁴

Conclusions about the impact of diagnosis on celiac patients and on their social environment demonstrates clearly the need to identify the persons involved in this process. The answer to the announcement of a chronic condition is processed differently both in mothers' and in the childrens' mind. While the former worries because she understands the term "chronicity", for the latter it is no more than a short- or very short-term matter. This is the reason for the child's serene behavior and for the mother's anguish and alarm. The child can quickly accept his or her condition since the matter will be completely handled by his or her parents or some other significant adult or trusted person in the family.

It is necessary to establish a communication protocol with the medical team, so as to fine-tune, each time more accurately, its influence in the management of the therapeutic process' course, of a better and fuller acceptance of the celiac condition and on a prompter and firmer acceptance of said condition.

The commercial outlet choice where to purchase or consume suitable dietary products, has a connotation which is completely concordant with the celiac condition, that is to say, with the need to be a part of the community, to be able to meet neighbors and friends at the supermarket, of erasing, even if only symbolically, the barriers that separate the patient from those who tolerate gluten. Purchasing food at a drug store suggests, for the celiac condition, a condition of discrimination.

It can be concluded that this kind of surveys are necessary and useful since they may afford a deeper understanding of celiac disease and of its social and emotional aspects.

References

1. Cueto-Rúa EA, Pecotche G. *La Enfermedad celíaca y su entorno. Creación del Club de Madres*. En: XI Congreso Argentino de Pediatría. Argentina, Mar del Plata. Sesión Temas Libres. Coordinador R. Maggi. 1981.
2. Drut R, Cueto-Rúa EA. *Análisis cuantitativo e inmunohistoquímico de la mucosa yeyunal de niños con enfermedad celíaca y con dieta libre de gluten*. Arch Argen Pediatr. 1985; 83: 20-4.
3. Comité de Gastroenterología de la Sociedad Argentina de Pediatría. *Conclusiones de la Jornada de Diagnóstico de Intestino Delgado*. Hospital de Niños de La Plata. Arch Arg Pediatr. 1986; 84: 38-9.
4. Cueto-Rúa EA, Menna ME, Morales V, Pecotche G. *Enfermedad celíaca y anticuerpos anti músculo liso*. Arch Arg Pediatr. 1986; 84: 269-73.
5. Cueto-Rúa EA, Menna ME, Morales V, Drut R. *Anticuerpos antimúsculo liso en la detección y seguimiento del enfermo celíaco*. Acta Gastroent Latinoamer. 1987; 3: 227-34.
6. Cueto-Rúa EA. *Enfermedad Celíaca*. Programa Nacional de Actualización Pediátrica 2004; 1: 13-30.
7. Cueto-Rúa EA, Gómez JC, Crivelli A, Guzmán L. *Guías, criterios y score de Diagnóstico*. Ministerio de Salud de la Poviaiciade Buenos Aires Disponible en: <http://www.ms.gba.gov.ar/EducacionSalud/celiaquia/celiaquia.html>
8. Programa de Enfermedad Celíaca. Ministerio de Salud de la Nación. Argentina. Disponible en: <http://www.msal.gov.ar/celiacos/w-criterios-biopsia.html>
9. Guzmán L, Cueto-Rúa EA. *Enfermedad celíaca*. Factores que influyen en la adherencia al tratamiento. Revista Ludovica Pediátrica 2011; 5: 166-74.
10. Gomez JC, Selvaggio GS, Viola M et al. *Prevalence of celiac disease in Argentina: screening of an adult population in the La Plata area*. Am J Gastroenterol. 2001; 96: 2700-04. <http://dx.doi.org/10.1111/j.1572-0241.2001.04124.x>
11. Mora M, Litwin N, Tocca MDC. *Prevalencia de enfermedad celíaca: estudio multicéntrico en población pediátrica en cinco distritos urbanos de Argentina*. Rev Argent Salud Pública. 2010; 1(4): 26-31. Disponible en <http://bit.ly/11iSAvB>
12. Dicke WK. *Coeliac disease. Investigation of harmful effects of certain types of cereal on patients with coeliac disease*. MD Thesis Univ Utrecht.
13. Dicke WK, Weijers HA, Van de Kamer JH. *Coeliac disease presence in weath of a factors faving feleterius effects in cases of coeliac diseas*. Acta Paediat. 1953; 42: 34-42. <http://dx.doi.org/10.1111/j.1651-2227.1953.tb05563.x>
14. Janatuinen EK, Pikkarainen PH, Kempainen TA, et al. *A comparison of diets with and without oats in adults with celiac disease*. N Engl J Med. 1995; 333: 1033-7. <http://dx.doi.org/10.1056/NEJM199510193331602>
15. Comino I, Real A, de Lorenzo L, Cornell H, López-Casado MÁ, Barro F, et al. *Diversity in oat potential immunogenicity: basis for the selection of oat varieties with no toxicity in coeliac disease*. Gut. 2011; 60: 915-22. <http://dx.doi.org/10.1136/gut.2010.225268>
16. Real A, Comino I, de Lorenzo L, Merchaán F, Gil-Humanes J, Giménez MJ, et al. *Molecular and immunological characterization of gluten proteins isolated from oat cultivars that differ in toxicity for celiac disease*. PLoS One. 2012; 7(12): e48365. <http://dx.doi.org/10.1371/journal.pone.0048365>

17. Edwards G, Leffler DA, Dennis MD, Franko DL, Blom-Hoffman J, et al. *Psychological correlates of gluten-free diet adherence in adults with celiac disease*. J Clin Gastroenterol. 2009; 43: 301-6. <http://dx.doi.org/10.1097/MCG.0b013e31816a8c9b>
18. Ruiz A, Polanco I. *Exposición al gluten y aparición de enfermedades autoinmunes en la enfermedad celíaca*. Pediatrka. 2002; 22: 311-9.
19. Hall NJ, Rubin G, Charnock A. *Source school of applied sciences, University of Sunderland, Sunderland, UK. Systematic review: adherence to a gluten-free diet in adult patients with coeliac disease*. Aliment Pharmacol Ther. 2009; 30: 315-30. <http://dx.doi.org/10.1111/j.1365-2036.2009.04053.x>
20. Kurppa K, Lauronen O, Collin P, et al. *Factors Associated with Dietary Adherence in Celiac Disease: A Nationwide Study*. Digestion. 2012; 86: 309-14. <http://dx.doi.org/10.1159/000341416>
21. Roma E, Roubani A, Kolia E, Panayiotou J, Zellos A, Syriopoulou VP. *Dietary compliance and life style of children with coeliac disease*. J Hum Nutr Diet. 2010; 23: 176-82. <http://dx.doi.org/10.1111/j.1365-277X.2009.01036.x>
22. Olsson C, Hörnell A, Ivarsson A, Sydner YM. *The everyday life of adolescent coeliacs: issues of importance for compliance with the gluten-free diet*. J Hum Nutr Diet. 2008; 21: 359-67. <http://dx.doi.org/10.1111/j.1365-277X.2008.00867.x>
23. Cueto-Rúa EA. *Estreñimiento. Una epidemia programada*. Revista Gastrohnp. 2011; 13: 58-65.
24. Cueto-Rúa EA, Miculán S. *Constipación y encopresis*. Ludovica Pediátrica. 2008; 2: 71-3.
25. Black JL, Orfila C. *Impact of coeliac disease on dietary habits and quality of life*. J Hum Nutr Diet. 2011; 24(6): 582-7. <http://dx.doi.org/10.1111/j.1365-277X.2011.01170.x>
26. Leffler DA, Edwards-George J, Dennis M, Schuppan D, Cook F, Franko DL, et al. *Factors that influence adherence to a gluten-free diet in adults with celiac disease*. Dig Dis Sci. 2008; 53: 1573-81. <http://dx.doi.org/10.1007/s10620-007-0055-3>
27. Errichiello S, Esposito O, Di Mase R, Camarca ME, Natale C, Limongelli MG, et al. *Celiac disease: predictors of compliance with a gluten-free diet in adolescents and young adults*. J Pediatr Gastroenterol Nutr. 2010; 50: 54-60. <http://dx.doi.org/10.1097/MPG.0b013e31819de82a>
28. Samaşca G, Iancu M, Pop T, Butnariu A, Andreica M, Cristea V, et al. *Importance of the educational environment in the evolution of celiac disease*. Labmedicine. 2011; 42: 497-501. <http://dx.doi.org/10.1309/LM1HFQPSN56ZQYLJ>
29. Hopman EG, Koopman HM, Wit JM, Mearin ML. *Dietary compliance and health-related quality of life in patients with coeliac disease*. Eur J Gastroenterol Hepatol. 2009; 21: 1056-61. <http://dx.doi.org/10.1097/MEG.0b013e3283267941>
30. Rosén A, Ivarsson A, Nordyke K, Karlsson E, Carlsson A, Danielsson L, et al. *Balancing health benefits and social sacrifices: A qualitative study of how screening-detected celiac disease impacts adolescents' quality of life*. BMC Pediatrics. 2011; 11: 32. <http://dx.doi.org/10.1186/1471-2431-11-32>
31. Moreno Rodríguez MA. *El arte y la ciencia del diagnóstico médico. Principios seculares y problemas actuales*. La Habana: Editorial. Científico-Técnica; 2001. P. 59.
32. Rancich AM, Gelpi RJ. *Análisis de los principios éticos en Juramentos Médicos utilizados en las Facultades de Medicina de la Argentina en relación al Hipocrático*. Medicina Buenos Aires. 1998; 58: 147-52.
33. Lain Entralgo P. *La relación médico enfermo*. Madrid: Revista de Occidente; 1964.

34. Hernández J. *Consejos de Martín Fierro y el viejo Vizcacha*. Disponible en:
<http://www.redargentina.com/refranes/consejosferro.asp>