

Life-threatening onset of coeliac disease: a case report and literature review

Matteo Guarino,¹ Edoardo Gambuti,¹ Franco Alfano,¹ Andrea Strada,² Rachele Ciccocioppo ,³ Lisa Lungaro,¹ Giorgio Zoli,¹ Umberto Volta,⁴ Roberto De Giorgio,¹ Giacomo Caio ^{1,5}

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¹Department of Morphology, Surgery and Experimental Medicine, University of Ferrara, Ferrara, Italy

²Department of Emergency Medicine, St. Anna University Hospital, Ferrara, Italy

³Department of Medicine, A.O.U.I. Policlinico G.B. Rossi and University of Verona, Verona, Italy

⁴Department of Medical and Surgical Sciences, University of Bologna, Bologna, Italy

⁵Celiac Center and Mucosal Immunology and Biology Research Center, Massachusetts General Hospital - Harvard Medical School, Boston, MA, United States

Correspondence to

Dr Giacomo Caio;
caigmp@unife.it

ABSTRACT

Background Coeliac disease (CD) results from an immune-mediated reaction to gluten in genetically predisposed individuals. In rare cases CD may occur with acute features deferring the diagnosis and exposing these patients to possible life-threatening complications. Herein we present the case of a young woman with a coeliac crisis, that is, a sudden clinical onset characterised by severe electrolyte imbalance due to an unknown (previously unrecognised) CD.

Methods This is a case report and literature review revealing that coeliac crisis is under-reported, with a total of 48 adult cases so far published. The diagnosis in our case was established by histopathological analysis of multiple duodenal biopsies. The patient's serum was tested by enzyme-linked immunoassay to detect antitransglutaminase IgA antibodies.

Results In contrast to cases reported in the literature, with male gender predominance and a mean age of 50±17 years, our patient was a young female case of coeliac crisis. However, like in our patient, a higher incidence of coeliac crisis was associated with the human leucocyte antigen (HLA)-DQ2 haplotype, versus HLA-DQ8, and a severe (Marsh-Oberhuber 3c) duodenal mucosa atrophy. Notably, there is no clear correlation between the antitissue transglutaminase 2 IgA antibody titre and coeliac crisis onset/severity, as confirmed by our case report.

Conclusions The present case highlights that CD may manifest quite abruptly with a severe malabsorption syndrome, that is, electrolyte abnormalities and hypoproteinaemia. Our case should alert physicians, in particular those in the emergency setting, that even a typically chronic disorder, such as CD, may show life-threatening complications requiring urgent management.

INTRODUCTION

Coeliac disease (CD) is a multisystemic, immune-mediated illness evoked by gluten ingestion in genetically susceptible individuals.¹ The main target organ of the auto-immune reaction against the enzyme tissue transglutaminase (TG2) is the small bowel, where the gluten-related inflammatory

cascade causes a progressive mucosal damage leading to severe villous atrophy.^{1 2} From a clinical standpoint, CD is a multifaceted chronic condition displaying a broad spectrum of intestinal (ranging from mild irritable bowel syndrome-like to severe malabsorption symptoms) and extraintestinal manifestations targeting several tissues and organs (eg, skin, endocrine/exocrine glands, nervous system, joint/muscles). As a result, CD remains a challenging condition to be diagnosed, thus causing a significant delay in establishing the appropriate therapy and increasing related morbidity.³⁻⁵

A potentially life-threatening and neglected clinical manifestation of CD is the so-called 'coeliac crisis', characterised by acute, massive watery diarrhoea, severe dehydration and metabolic disturbances, leading to neuromuscular weakness, tetanic seizures, cardiac arrhythmias and even sudden death in extreme cases.⁶⁻⁸ This condition is largely under-reported and under-recognised both in children and adults, with a total of 48 adult cases published so far.⁶⁻⁴⁶ In most cases, coeliac crisis develops due to voluntary or inadvertent gluten ingestion in patients with or without an established diagnosis of CD. Only rarely a coeliac crisis heralds the clinical onset of CD, requiring hospitalisation and rapid therapeutic management due to possible occurrence of severe complications with high morbidity and mortality.⁹⁻¹³

Herein we describe the case of a patient admitted to our emergency department for a severe life-threatening coeliac crisis as the first manifestation of a previously unknown CD.

CASE REPORT

A 34-year-old woman was admitted to the emergency unit complaining of limb

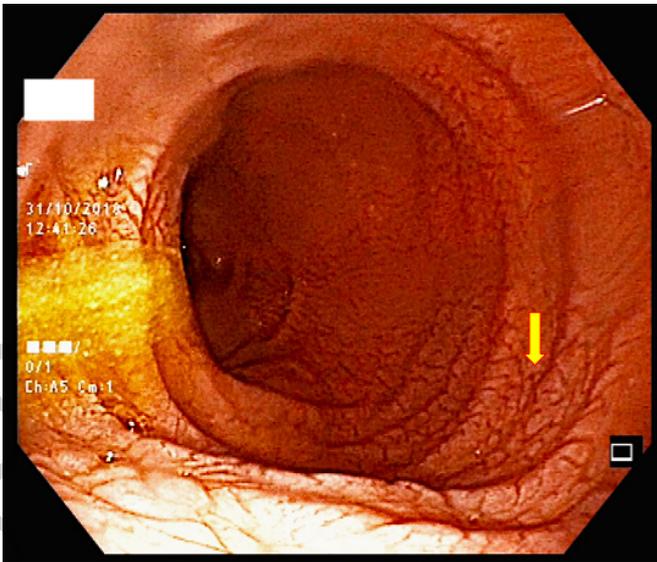


Figure 1 Representative duodenal endoscopic picture showing decreased mucosal folds carrying a scalloping profile, together with a mosaic pattern and an increased vascular network (arrow), all suggestive of villous atrophy.

numbness and watery diarrhoea (8–10 bowel movements/day) which started 2 weeks earlier. The patient reported a weight loss of about 10 kg in the last 2 months in the absence of hyporexia. Her clinical history unravelled microcytic anaemia treated with oral iron replacement. Physical examination showed severe weakness of the limbs with a bilaterally positive Trousseau's sign without cardiorespiratory abnormalities. Vital parameters were within the normal range. The abdomen was flat, without tenderness, while auscultation disclosed increased intestinal sounds. Her ECG showed a sinus rhythm with type 1 atrioventricular block, flat T waves associated with U waves and an elongated QTc interval

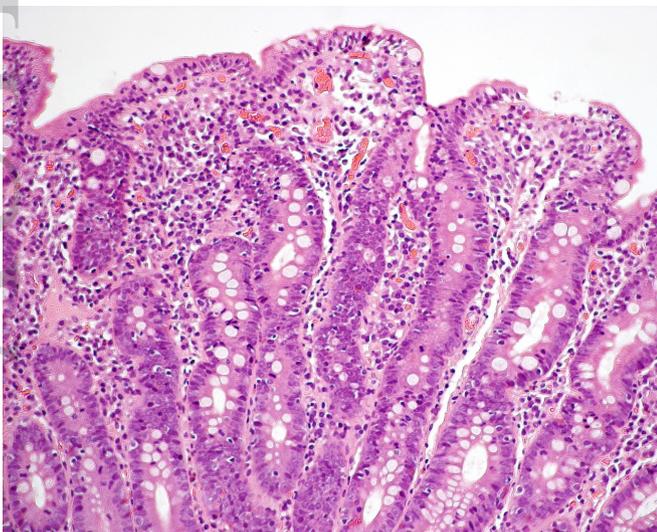


Figure 2 Representative microphotograph of the duodenal mucosa showing villous atrophy and crypt hyperplasia with dense inflammatory infiltrate of the lamina propria (Marsh-Oberhuber lesion grade 3c). H&E staining, original magnification 200 \times .

(570 ms). Laboratory tests revealed severe electrolyte imbalance, with hyponatraemia (133 mmol/L), hypokalaemia (1.6 mmol/L), hypocalcaemia (ionised calcium of 0.9 mmol/L), hypophosphataemia (1.6 mg/dL) and hypomagnesaemia (1.4 mmol/L). Furthermore, the patient had hypochromic microcytic anaemia (haemoglobin of 85 g/L, with a mean cell volume of 68 fL and a mean cell haemoglobin of 20.6 pg), normal platelet count ($297 \times 10^9/L$), iron (serum iron 18 $\mu\text{g}/\text{dL}$; ferritin 2 ng/mL) and folate deficiency (2 ng/mL), as well as hypoproteinaemia and hypoalbuminaemia (total serum protein 4.4 g/dL; albumin 2.6 g/dL). Due to severe electrolyte imbalance, a conspicuous electrolyte replacement was rapidly administered, leading to a slight improvement in electrocardiographic abnormalities. The patient was then admitted to the internal medicine ward for adequate investigation and treatment. During the hospitalisation, the common causes of infectious diarrhoea were excluded by stool cultures, and the faecal occult blood test resulted negative. Both ultrasound and abdominal X-ray examinations were unremarkable. Liver function tests revealed a slight increase of transaminases, with alanine transaminase and aspartate transaminase values of 47 U/L and 60 U/L (n.v. 5–35 for both parameters), respectively. Based on the lack of fever, normal C reactive protein and the presence of non-bloody, watery diarrhoea, the patient was further evaluated with an upper endoscopy. The examination revealed stigmata of villous atrophy at the duodenal level (figure 1), where biopsies were taken from the bulb and the second portion. Histopathological analysis showed the presence of a severe villous atrophy (Marsh-Oberhuber grade 3c) (figure 2) without any evidence of aberrant intraepithelial lymphocytes.¹⁴ Based on the histopathological result, we used enzyme-linked immunoassay to test IgA anti-TG2, which turned to be positive at low titre (23 U/mL, n.v. <10 U/mL). This result was associated with the positivity of IgA antiendomysial antibodies (1:80) revealed by indirect immunofluorescence.^{47 48} The genetic test showed human leucocyte antigen (HLA)-DQ2 positivity. Therefore, a firm diagnosis of CD was established and the patient started a gluten-free diet (GFD). Due to rapid improvement after gluten withdrawal, a course with steroid treatment was deemed not necessary. Since diarrhoea and paraesthesia showed significant improvement with complete regression in about a week, the patient was discharged in good health.

DISCUSSION AND REVIEW OF THE LITERATURE

In the vast majority of cases, the natural history of CD is characterised by chronic evolution without acute exacerbations. Conversely, coeliac crisis is burdened by severe acute symptoms such as abdominal pain and distension, massive diarrhoea and weight loss, causing a life-threatening malabsorption syndrome. In most cases, gluten is introduced inadvertently, whereas in some patients with poor compliance to GFD a voluntary

Table 1 Synopsis highlighting the main features of adult cases (n=48) with coeliac crisis published so far

Reference	Diagnosis														
	Age	Sex	Weight loss	Favorable evolution with GFD	Biopsy (Marsh grade)	EMA	TG2 (UI/mL)	HLA	Hb (g/L)	PLT ($\times 10^9/L$)	INR	Hypo-proteinaemia	Electrolyte Abnormalities	LOS (days)	Death
Ozslan <i>et al</i> ⁸	75	M	Yes	Yes	N/A	Positive	N/A	N/A	59	97	N/A	Yes	Yes	10	No
Poudyal <i>et al</i> ¹⁰	55	F	N/A	Yes	N/A	Positive	N/A	N/A	79	43	N/A	Yes	Yes	8	No
Gutiérrez <i>et al</i> ¹¹	20	M	No	Yes	N/A	N/A	100	DQ8	86	N/A	N/A	No	Yes	N/A	No
Wolf <i>et al</i> ¹²	26	F	N/A	Yes	3c	N/A	23.4	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
Forrest <i>et al</i> ¹⁵	36	F	Yes	Yes	3c	N/A	N/A	N/A	<110	N/A	N/A	N/A	Yes	N/A	No
Akbal <i>et al</i> ¹⁶	43	F	Yes	Yes	3c	N/A	608	N/A	78	<100	2.1	Yes	Yes	107	No
Al Shammeri <i>et al</i> ¹⁷	52	F	Yes	Yes	N/A	Positive	Positive	N/A	<100	N/A	N/A	Yes	Yes	N/A	No
Atikou <i>et al</i> ¹⁹	50	F	N/A	Yes	N/A	1/160	15	N/A	N/A	N/A	N/A	N/A	Yes	N/A	No
Parry and Acharya ²⁰	26	F	N/A	Yes	3	Positive	N/A	N/A	N/A	N/A	N/A	No	Yes	N/A	No
Magro and Pullicino ²¹	83	M	N/A	Yes	N/A	Positive	6	N/A	N/A	N/A	N/A	N/A	N/A	N/A	No
Lindo Ricce <i>et al</i> ²²	38	M	Yes	Yes	3c	N/A	35.9	N/A	152	N/A	1.6	Yes	Yes	N/A	No
de Almeida Menezes <i>et al</i> ²⁵	63	F	N/A	No	3c	N/A	45	DQ2	N/A	N/A	N/A	Yes	Yes	30	No
Gonzalez <i>et al</i> ²⁶	31	M	Yes	Yes	N/A	N/A	N/A	N/A	N/A	N/A	3.48	Yes	Yes	6	No
Da Costa Becker <i>et al</i> ²⁷	76	M	Yes	Yes	N/A	N/A	N/A	N/A	102	N/A	>10	N/A	Yes	10	No
Ferreira <i>et al</i> ²⁸	49	F	Yes	Yes	3c	Positive	Positive	N/A	72	250	>1.5	Yes	Yes	N/A	No
Helvaci <i>et al</i> ²⁹	56	M	Yes	Yes	N/A	N/A	N/A	DQ2	<110	N/A	>1.2	No	Yes	7	No
Chen <i>et al</i> ³⁰	25	F	Yes	Yes	3c	N/A	100	N/A	N/A	N/A	N/A	Yes	Yes	14	No
Sbai <i>et al</i> ³¹	24	F	Yes	Yes	3c	N/A	99	N/A	93	N/A	3.2	No	Yes	8	No
Bul <i>et al</i> ³²	43	M	Yes	Yes	3c	N/A	19	N/A	N/A	N/A	N/A	No	Yes	5	No
Tiwari <i>et al</i> ³³	46	M	Yes	Yes	N/A	N/A	48	N/A	110	N/A	N/A	Yes	No	N/A	No
Bou-Abboud <i>et al</i> ³⁴	26	F	N/A	Yes	N/A	N/A	100	N/A	158	N/A	N/A	Yes	Yes	N/A	No
Yilmaz <i>et al</i> ³⁵	69	M	N/A	Yes	3b	N/A	132	N/A	48	N/A	1.2	Yes	Yes	N/A	No
Ribeiro do Vale <i>et al</i> ³⁶	75	M	No	Yes	N/A	3+	Positive	N/A	81	180	N/A	Yes	Yes	6	No
Mrad <i>et al</i> ³⁷	82	F	Yes	N/A	N/A	1+	Positive	N/A	76	156	N/A	Yes	Yes	8	No
	58	M	Yes	Yes	3c	1/640	142	N/A	116	N/A	1.6	No	Yes	11	No
	64	F	Yes	Yes	3c	Positive	200	N/A	87	N/A	>1.2	Yes	Yes	N/A	No

Continued

Table 1 Continued

Reference	Age	Sex	Weight loss	Favorable evolution with GFD	Diagnosis		EMA	TG2 (UI/mL)	HLA	Hb (g/L)	PLT (x10 ⁹ /L)	INR	Hypo-proteinaemia	Electrolyte Abnormalities	LOS (days)	Death
					Biopsy (Marsh grade)	Biopsy (Marsh grade)										
Jamma <i>et al</i> ³⁸	34	F	Yes	Yes	3b	N/A	113	DQ8	N/A	N/A	N/A	N/A	N/A	Yes	7	No
	51	M	Yes	Yes	3c	N/A	200	DQ2	N/A	N/A	N/A	N/A	N/A	Yes	11	No
	48	F	Yes	Yes	3b	N/A	0	DQ2	N/A	N/A	N/A	N/A	N/A	Yes	4	No
	70	M	Yes	Yes	3	N/A	100	N/A	N/A	N/A	N/A	N/A	N/A	Yes	N/A	No
	48	F	Yes	N/A	3	N/A	N/A	N/A	DQ2	N/A	N/A	N/A	N/A	Yes	7	No
	68	F	Yes	Yes	3	N/A	6	N/A	DQ2	N/A	N/A	N/A	N/A	Yes	5	No
	67	F	Yes	Yes	3c	N/A	250	DQ2	N/A	N/A	N/A	N/A	N/A	Yes	8	No
	74	F	Yes	Yes	3c	N/A	24.5	N/A	N/A	N/A	N/A	N/A	N/A	No	7	No
	65	M	Yes	Yes	3	N/A	21.3	DQ2	N/A	N/A	N/A	N/A	N/A	Yes	10	No
	68	M	Yes	Yes	3b	N/A	117	DQ2	N/A	N/A	N/A	N/A	N/A	Yes	11	No
	65	F	Yes	Yes	3c	N/A	22	N/A	DQ2	N/A	N/A	N/A	N/A	No	13	No
	49	F	Yes	Yes	3	N/A	83.7	N/A	DQ2	N/A	N/A	N/A	N/A	Yes	4	No
	Hammami <i>et al</i> ³⁹	26	F	Yes	No	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	Yes	7	Yes
	Toyoshima <i>et al</i> ⁴⁰	30	F	Yes	Yes	N/A	1/1280	5	N/A	114	N/A	N/A	N/A	Yes	N/A	No
Kelly <i>et al</i> ⁴¹	23	F	N/A	Yes	N/A	N/A	33.9	N/A	120	94	N/A	N/A	Yes	21	No	
Krishna <i>et al</i> ⁴²	67	M	Yes	Yes	3	Negative	4	DQ8	124	160	N/A	N/A	Yes	15	No	
Chandan <i>et al</i> ⁴³	58	M	Yes	Yes	3c	N/A	200	N/A	47	N/A	N/A	N/A	Yes	N/A	No	
Kizilgul <i>et al</i> ⁴⁴	50	M	Yes	Yes	3	158.7	200	N/A	94	N/A	N/A	N/A	Yes	N/A	No	
Selen <i>et al</i> ⁴⁵	37	F	Yes	Yes	3	Positive	Positive	N/A	80	N/A	N/A	N/A	Yes	N/A	No	
Gupta <i>et al</i> ⁴⁶	30	F	Yes	Yes	3b	N/A	N/A	N/A	<110	N/A	N/A	N/A	Yes	7	No	
Present Case	34	F	Yes	Yes	3c	1/80	23	DQ2	85	297	2.52	Yes	Yes	9	No	

EMA, antiendomysial antibodies; F, female; GFD, gluten-free diet; Hb, haemoglobin; HLA, human leucocyte antigen; INR, international normalised ratio; LOS, length of stay; M, male; N/A, not applicable; PLT, platelets; TG2, tissue transglutaminase.

ingestion may occur.^{3 6 7} Only seldom a coeliac crisis can herald the onset of CD,¹⁰ as it was in the herein reported case. Among the reported cases described in table 1, there was a greater prevalence of female than male gender (28 vs 20), with a mean age of 50±17 years, which is higher than the age of the present case.^{9–46} Like in the present case, a higher incidence of coeliac crisis has been reported in patients carrying the HLA-DQ2 haplotype of genetic susceptibility to the disease as compared with those with HLA-DQ8, whereas the biopsy specimens showed signs characteristic of Marsh-Oberhuber 3 (from ‘a’ to ‘c’) mucosal lesions.¹ Other common clinical features included weight loss, hypoproteinaemia and electrolyte abnormalities, whereas coagulation abnormalities (ie, prothrombin time elongation) along with markedly reduced platelet count were uncommon. No clear correlation was found between the anti-TG2 IgA antibody titre and coeliac crisis onset/severity, as also supported by our case showing only a twofold increase above the upper normal limit.

Among the major features of this case were the abrupt onset of symptoms, in particular of those related to electrolyte imbalance, and the severity of the clinical picture prompting admission to the emergency department. Further to severe dehydration, the patient presented with neuromuscular weakness, a finding detected in other reports,^{7 9 13–21} and electrocardiographic abnormalities due to the key role exerted by potassium in regulating cell excitability. Likely, hypocalcaemia and hypomagnesaemia also contributed to worsening cellular excitability. According to previous evidence,^{6 9–13} hypoproteinaemia with hypalbuminaemia and metabolic acidosis were found in our patient as hallmarks of malabsorption, whereas paraesthesia was likely related to electrolyte imbalance. However, the variety of possible clinical pictures in coeliac crisis should be underlined, ranging from central nervous system involvement with tetraplegia/paraplegia and ataxia,^{17–19} psychosis²² as well as seizures,^{23 24} to coagulopathy^{11 25–27} and acute kidney injury.^{28 29} In all cases described so far, coeliac crisis required urgent hospitalisation. The milestone treatment is fluid resuscitation with correction of the electrolyte imbalance, which can lead to life-threatening cardiac arrhythmias. Nutritional support is also of paramount importance, and clinicians should take coeliac crisis in mind during differential diagnosis of severe acute diarrhoea with weight loss, as patients’ prognosis can dramatically improve with a simple dietary intervention.

In our case, like in almost all cases described, GFD led to a dramatic improvement in clinical picture. Nutritional management should take into consideration the possible occurrence of a refeeding syndrome, which can be fatal if not recognised and treated properly, as described in one patient with coeliac crisis.³⁹ In less than 20% of cases (8 of 48 cases, 16%), corticosteroids were administered during management.⁴⁹ However, we decided not to use steroids since their usefulness was recently questioned.^{38 50} It was reported previously that immunosuppression with

corticosteroids and azathioprine for autoimmune hepatitis or prednisone for Bell’s palsy did not prevent the occurrence of coeliac crisis in patients.^{17 51} Moreover, steroid therapy may increase electrolyte depletion, facilitating the occurrence of refeeding syndrome.²² Finally, despite the acute onset of malabsorption syndrome in adulthood, our case did not show features of complicated CD^{52 53} (ie, refractory CD), and the clinical picture dramatically improved in a few days with GFD, still the only effective treatment available.⁵⁴

CONCLUSION

The present case highlights the possibility that CD may manifest quite abruptly with a severe malabsorption syndrome and related electrolyte abnormalities and hypoproteinaemia. This would imply that even a typically chronic disorder, such as CD, may have an acute onset in a small proportion of patients, which emergency physicians should be aware of. Although rarely encountered in clinical practice, this acute onset of CD requires hospitalisation and immediate treatment (ie, electrolyte replacement and protein correction) in order to avoid life-threatening complications.

Contributors MG, RDG and GC: design and conceptualisation. MG, EG and FA reviewed the literature and wrote the first draft of the manuscript. AS, RC, LL, GZ, UV, RDG and GC participated in the clinical assessment of the patient and critically reviewed the paper.

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ORCID iDs

Rachele Ciccocioppo <http://orcid.org/0000-0002-6297-7576>

Giacomo Caio <http://orcid.org/0000-0002-4244-4529>

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