

## CASE REPORT

# Celiac Crisis Presenting as Status Epilepticus and Irreversible Encephalopathy: A Rare Pediatric Case Report

SAADIA KHAN, SUMMERA TABASUM, REEMA ARSHAD, ASAD ABBAS, TEHSEEN IKRAM, NAZIA BATOOL

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### ABSTRACT

#### Correspondence to:

**Reema Arshad,**  
Clinical Nutritionist,  
Department of Preventive  
Pediatrics, Children Hospital and  
Institute of Child Health, Multan

**E-mail:** reemaarshad0@gmail.com

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Celiac disease is an immune-mediated enteropathy manifested as malabsorption, diarrhea, and failure to thrive, with symptoms as dehydration, electrolyte imbalance, metabolic derangements. Along with neurological symptoms, the first presentation as status epilepticus and subsequent irreversible encephalopathy is extremely rare. The present case is a 2.5 years old female presented to the emergency department with status epilepticus and encephalopathy. The patient was in usual state of health prior to illness at home with no history of abdominal distension, intermittent diarrhea. The patient presented to tertiary care in emergency with status epilepticus and encephalopathy with probable diagnosis of tuberculous meningitis (TBM). And then referred to the Children Hospital and the Institute of Child Health Multan for status epilepticus at the emergency department. In the context of this case, it is suggested that celiac crises should be considered in the differentials of seizures and encephalopathy.

**Key Words:** *Celiac crisis, Celiac disease, Encephalopathy, Global involution, Irreversible encephalopathy, Pediatric, Status epilepticus*

### INTRODUCTION

Celiac disease (CD) is a gluten sensitive enteropathy of small bowel characterized by villous atrophy that leads to malabsorption steatorrhea and weight loss.<sup>1,2</sup> A broad spectrum of disorders, especially autoimmune, is associated with celiac disease ranging from biliary cirrhosis, Sjogren's syndrome to dermatitis herpetiformis. The most common presentation is gastrointestinal malabsorption symptoms.<sup>3,4</sup>

The neurological symptoms of CD are ataxia, epilepsy, neuropathy, and myelopathy.<sup>3-7</sup> The causes are still not clear, but it is hypothesized that the pathogenesis of this relation is either by the deficiencies of the micronutrients (as Vitamins

B12, D or E), autoimmunity, hereditary or gluten toxicity.<sup>4,8</sup>

Celiac crisis is a rare life-threatening manifestation of celiac disease presented mostly with severe dehydration and shock, severe metabolic imbalance, including loss of potassium, calcium and metabolic acidosis.<sup>9,10</sup> Neurological symptoms of celiac crisis i.e. that of status epilepticus and encephalopathy are extremely rare with limited published data.<sup>9</sup> The development of a crisis, with possibly fatal, central nervous system (CNS) complications, rather than gastrointestinal symptom is not well known.<sup>6,9</sup>

The present case itself is quite rare with no local case reported. Hence, the case of the celiac crisis

in the pediatric patient with significant signs of the status epilepticus and incurable encephalopathy are presented in the current report.

### CASE REPORT

**Clinical Course and Management:** A 2.5-year-old female child belonging to a low socio-economic background presented with an acute illness. Her prior history at home was unremarkable, with no history of abdominal distension or intermittent diarrhea. She was brought to a tertiary care emergency department with status epilepticus and encephalopathy, with a probable diagnosis of tuberculous meningitis. The patient was subsequently referred to the Children's Hospital & Institute of Child Health for further management. There was no history of contact with tuberculosis, and she was vaccinated according to the Expanded Programme on Immunization schedule. Her developmental history was appropriate for age.

**Presentation:** The sick child presented with generalized seizures, severe dehydration with metabolic acidosis and GCS 9/15. She was pale, emaciated and wasted with weight for height <-4 SD. Clubbing was present. There was soft abdomen with sluggish bowel sounds. Neurological examination showed GCS of 9/15 with generalized increased tone with preserved cranial nerves and sensory functions. Signs of meningeal irritation were absent. Fundoscopy showed no sign of raised Intracranial pressure.

1. **Emergency stabilization:** Patient was stabilized initially at the emergency department as per protocol for status epilepticus with proper monitoring. After initial stabilization of seizures fluid resuscitation, correction of metabolic acidosis, and treatment of complications along with proper diagnostic workup were carried out.
2. **Treatment:** The patient received antiepileptic therapy, intravenous fluids, electrolyte correction, calcium, potassium, micronutrients, antibiotics, and intravenous steroids were administered.
3. **Clinical labs:** There was microcytic hypochromic anemia 8.1 g/dl, hyponatremia 128.mm/lit, hypoglycemia 2.5 mmol/lit and hypokalemia 2.8 mmol/lit, EEG revealed

diffuse slowing of background activity no epileptogenic activity seen. Initial MRI brain was normal but repeat imaging showed global involuntional changes. Fundoscopy showed no signs of raised ICP in work up of TBM, PPA scoring was not significant on infectious diseases screening HBV HCV Syphilis and ICT malaria were negative.

- The **Cerebrospinal fluid (CSF)** was sterile and staining for TBM was negative. On culture and microscopy no atypical or malignant cells were seen.
4. ABGs showed metabolic acidosis with PH 7.2, BE -14, CO2 30 mmHg, serum albumin 2.4 g/dl, creatinine 0.4 mmol/lit. NH<sub>4</sub> and lactate were normal (NH<sub>4</sub>=40, lactate=15). Anti-tissue transglutaminase IgG was 320 IU/lit.
  5. **Nutrition Rehabilitation:** NG feed was introduced which was gradually substituted with 220 Kcal/kg /day feed, introduction of the micronutrient supplements and a course of intravenous administration of the methylprednisolone will ( 2mg/kg/day over 5 days) was given as a life-saving intervention.
  6. **Outcome:** Even though metabolic issues were resolved, the neurological outcome was unfavorable. The girl was still depressed, irritated and very sick and encephalopathic. An assessment of a brain MRI after a follow up revealed that there was extensive global atrophy which is irreversible as far as a brain is concerned.

### DISCUSSION

Celiac crisis is the most severe and life-threatening form of celiac disease, leading to severe dehydration, shock, electrolyte imbalance, and profound metabolic acidosis.<sup>7,10</sup> It classically presents in children, particularly those from low socio-economic backgrounds; however, more recent evidence indicates that it can occur at any age and may be triggered by infections, metabolic stress, or sudden exposure to gluten. Early diagnosis and prompt management are mandatory and are associated with reduced mortality and morbidity.<sup>11</sup> Central nervous system (CNS) manifestations of celiac crisis are extremely rare in children with celiac disease. Cerebellar ataxia, epilepsy, peripheral

neuropathy, and myopathy are the most commonly reported neurological manifestations and usually present after infancy and beyond two years of age.<sup>9</sup> Other manifestations may include paralysis or quadriplegia secondary to hypokalemia, even in the absence of significant gastrointestinal symptoms. The exact pathophysiology of neurological involvement is not well understood; however, immunological, nutritional, toxic, and metabolic disturbances have been postulated as possible mechanism.<sup>12,13</sup> However, more evidence suggested that neurological signs in celiac diseases are more commonly caused by cross-reaction of antibodies, immunological process and direct gluten toxicity, along with vitamin B12 deficiency vitamin D deficiency and vitamin E deficiency.<sup>6</sup>

In our case, the patient developed a prolonged coma with irreversible encephalopathy, most likely related to gluten-mediated neurotoxicity, leading to cerebral involution. A similar case was reported by Basser et al., describing a two-year-old boy with reversible encephalopathy; however, in contrast, our patient was a 2.5-year-old female who developed irreversible encephalopathy.<sup>11</sup> Various neurologic manifestations have been reported in children encephalopathy and global involution is rarely documented.<sup>11,12</sup> Another case reported by Bashir et al reported celiac disease in 16-month-old child with motor regression which showed good neurodevelopmental outcome with gluten restriction.<sup>2</sup>

Delayed diagnosis is one of the significant causes of poor outcome especially in the South Asian population where chronic diarrhea and malnutrition are often associated with infections or lactose intolerance. Untreated long-term malabsorption leads to electrolyte imbalances, hypoalbuminemia, dehydration, and neurological alterations. These are critical to prevent multi-organ dysfunction.<sup>7</sup> Early Management requires rehydration, electrolytes balance, and pH stabilization. Ketosis and hypoglycemia must be dealt with through glucose infusion. It is highly advised that thiamine supplementation should be used to avoid Wernicke-like encephalopathy. In a gluten-free diet, the fixation should come after stabilization to prevent the aggravation of the metabolic imbalance.<sup>14,15</sup> In this example, the metabolic basis of encephalopathy is supported

by the fact that the patients improved after stabilization.

The case highlights the need to consider celiac disease as a cause of the chronic diarrhea and severe malnutrition among children in the high-burden countries like Pakistan. It further indicates that celiac crisis may manifest itself through encephalopathy and that early detection and therapy are essential to save lives. Enhancing screening pathways, education of caregivers and early referral of serology may play an important role in reducing severe presentations. The case contributes to the scanty regional literature and supports the necessity of the clinician training and new management of encephalopathy.

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#### Authors' affiliation

**Dr. Saadia Khan, MD,**  
Assistant Professor,  
Department of Preventive Pediatrics, Children Hospital and Institute of Child Health (CH & ICH, Multan),

**Dr. Summera Tabsum, MBBS,**  
General Medical Practitioner,  
Children Hospital and Institute of Child Health, Multan.

**Reema Arshad, MSc (Food and Nutrition),**  
Clinical Nutritionist,  
Department of Preventive Pediatrics, Children Hospital and Institute of Child Health (CH & ICH, Multan).

**Dr. Asad Abbas, MPhil Human Nutrition and Dietetics,**  
Assistant Professor,  
Multan College of Food and Nutrition, Multan.

**Dr. Tehseen Ikram, MBBS, WMO,**  
Student,  
Department of Preventive Pediatrics, Children Hospital and Institute of Child Health (CH & ICH, Multan),

**Nazia Batool, BS Honor, PGD,**  
Food and Nutrition,  
Clinical Nutritionist, TZP Trust,

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#### Authors' contribution

**SK:** Proposed topic, basic study design, methodology and manuscript writing

**ST:** Data collection, statistical analysis and interpretation of results etc.

**RA:** Literature review & referencing and quality insurer

**AA:** Data collection and interpretation

**TI:** Paper writing and final revision

**NB:** Statistical analysis

*All the authors have approved the final manuscript draft and accept the responsibility of research integrity.*