

Hikikomori, a risk factor for life-threatening malnutrition: a case report of an emergency within the vulnerability

Nicola Artusi, Emanuele Russo, Gianmaria Valeri, Marzia Tecchiolli, Franco Cominotto

Emergency Medicine Department, Azienda Sanitaria Universitaria Integrata di Trieste, Italy

Abstract

The Japanese term “hikikomori” represent a condition of self-isolation, predominantly seen in male adolescents and young adults. Hikikomori is associated with malnutrition, mood disorders, self-neglect, and reduced access to healthcare. Although initially observed and described in Japan, this psychiatric condition has become a global phenomenon in recent years, with an important burden on patients and their families. Hikikomori syndrome can be especially dangerous when compounded by psychiatric

comorbidities or socioeconomic vulnerabilities, as evidenced by an alarming increase during the COVID-19 pandemic. We present the case of a young man admitted to the emergency department with life-threatening malnutrition and anemia after prolonged self-isolation following cyberbullying episodes. The patient was found in critical condition, suffering from shock and multiorgan failure. After initial emergency interventions, and post-stabilization, he was transferred to psychiatric care. Emergency physicians should start considering hikikomori as a risk factor in the assessment of malnutrition and anaemia, above all in young adults. In addition, we want to highlight how much the COVID-19 pandemic exacerbated the critical issues of local medicine, exposing the most vulnerable to greater risk. The Emergency Department necessitates a more integrated approach with social and out-of-hospital health services for intercepting and addressing the needs of vulnerable individuals.

Correspondence: Nicola Artusi, Emergency Medicine Department, Azienda Sanitaria Universitaria Integrata di Trieste, Italy.
E-mail: nicola.artusi@asugi.sanita.fvg.it

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Introduction

The Japanese term “hikikomori” represents a condition of self-isolation, occurring mostly in male adolescents and young men.^{1,2} Its incidence worldwide has been alarmly increasing in recent years. Since 2020, the Sars-Cov-2 pandemic has aggravated social marginalization, especially among vulnerable populations, both in Italy and abroad;^{3,4} it also highlighted the criticalities of the local healthcare system, with the abandonment of part of the most vulnerable population, with strong repercussions on collective physical,⁵ psychological^{6,7} and social health.⁸

Proteinemia and hemoglobin (Hb) levels broadly provide an overview on patients' nutritional status. Anemia itself, above all, is a universal indicator of personal health and social integration.⁹ We present one case of life-threatening anemia in a young patient suffering from hikikomori syndrome admitted to an Italian emergency department in critical conditions after more than a year without any access to healthcare.

Case Report

In December 2021, a 24 year-old male was admitted to an Emergency Department; the emergency call started from the mother, who found the patient collapsed on the ground, at 2 a.m. The emergency responders found what they described as a “hoarding environment”; the patient was lying on the ground, in dire hygienic conditions, minimally responsive, extremely pale and cold to the touch; the mother claimed that her son suffered from a form of undefined psychosis and therefore he wouldn't communicate. At the arrival in the Emergency Department, the patient complained of abdominal pain. His past medical records were completely silent. The patient was severely underweight, cachectic, hypothermic with a core temperature of 34.0°C, hypotensive (90/50 mmHg), tachycardic and tachypnoeic. The abdomen was tender.

The point-of-care ultrasound showed a moderate left ventricle dysfunction, with diffused hypokinesia. An Arterial Blood Gas analysis (ABG) showed a moderate metabolic acidosis, pH 7.25, and a life-threatening Hb level of 1.8 g/dL. Immediate treatment involved multiple transfusions of 0 Rh-negative blood units, urgent intubation, noradrenaline infusion for hemodynamic support and external heating. The patient was admitted to the ICU, where minimal enteral nutrition was started, and he was repeatedly transfused. A contrast-enhanced Angio-CT scan was performed: although no source of bleeding was found, the CT scan reported bibasal pleural consolidation and effusion, severe periportal edema (Figures 1 and 2) and a 17 mm area of functional jejunal intussusception (Figure 3). A second CT scan, conducted the following day, after the initiation of blood transfusions and enteral nutrition, revealed complete remission of both the intussusception and hepatic edema. No sources of bleeding were identified once again.

Blood tests revealed acute liver failure with spontaneous INR 2.01, total bilirubin 4.64 mg/dL, creatinine 1.08 mg/dL, myoglobin 214.5 mcg/L (range <70), platelets $16 \times 10^3/\mu\text{L}$, LDH 3283 U/L (range <250), albumin 3.06 g/dL (range: 3.50-5.20), total proteins 4.6 g/dL (range 6.0-8.0), CRP 0.6 mg/L (range <5.0) in a global clinical presentation of shock and multiorgan failure with rhabdomyolysis.

The first hematocrit sample obtained in the Emergency Department was insufficient and coagulated due to the patient's critical state of shock and hypoperfusion; therefore, it could not be analyzed. The subsequent complete blood count, collected following the transfusion of five blood units, indicated the following values: WBC $16.97 \times 10^3/\mu\text{L}$; RBC $2.11 \times 10^6/\mu\text{L}$; Hb 6.3 g/dL; hematocrit 20.7%; MCV 98.1 fL; MCH 29.9 pg; MCHC 30.4 g/dL; RDW 16.3%; PLT $19 \times 10^3/\mu\text{L}$. Further laboratory examinations reported a panhypolipemia and panhypoprotidemia, associated with hypertransferrinemia and folate (2.3 ng/mL) and vitamin B12 (59 pg/mL) deficiencies in the global frame of severe malnutrition.

The CBC highlighted peculiar irregularities: the severe thrombocytopenia and the increased MCV and WBC count led to fur-

ther investigations including a bone marrow biopsy with immunophenotyping and subsequent hematological consult for exclusion of malignancy. A predominance for altered immature cells was noted, both in the granulopoietic and hematopoietic lineages, with poor representation of erythroblasts, which posed the indication for a further bone-marrow sampling and analysis, conclusive for chronic hematopoietic failure due to severe nutritional deficiency. After being successfully extubated, a dietary assessment was performed on the seventh day following hospital admis-

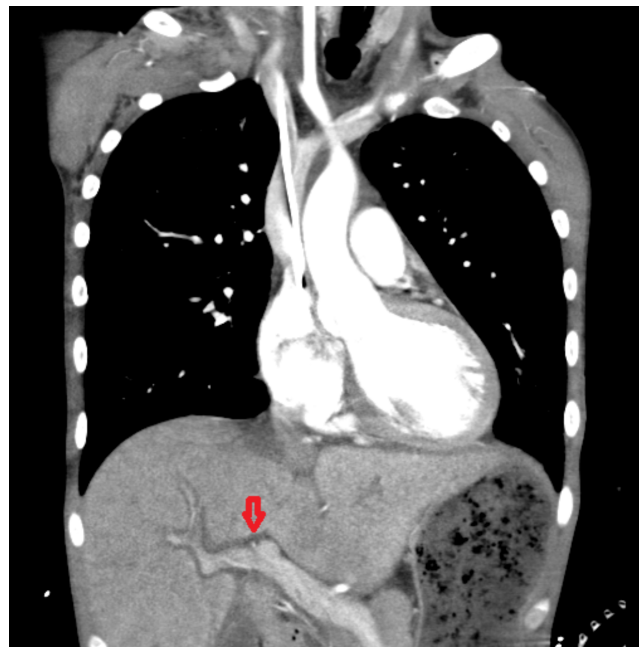


Figure 2. In red, the periportal edema in coronal projection.

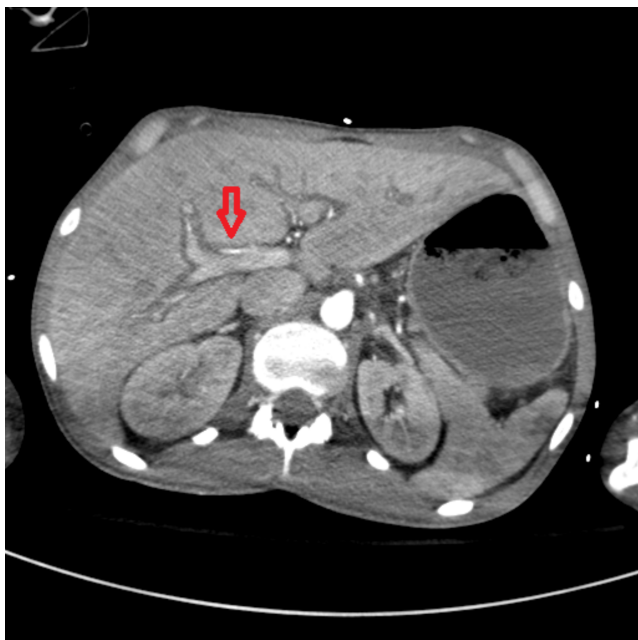


Figure 1. In red, the periportal edema in transversal projection.

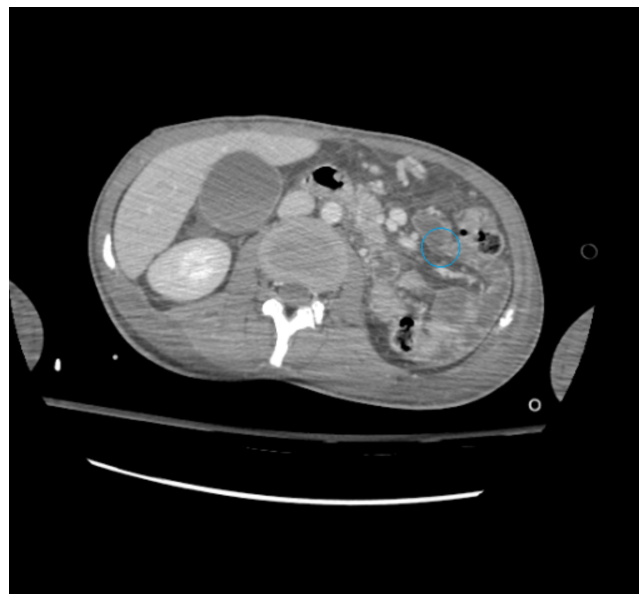


Figure 3. In blue, the small jejunal intussusception, expression of functional invagination.

sion. A light oral diet was initiated, along with the continuation of parenteral nutrition delivering 1070 kcal/1000 mL at an infusion rate of 50 mL/h over a 24-hour period. At the time of assessment, the patient weighed 51 kg with a height of 175 cm, corresponding to a BMI of 16.6 indicative of significant underweight.

During hospital stay, a permanent hearing loss was observed, and subsequently confirmed by audiometric assessment, attributed to a chronic vitamin deficiency.

The patient was later transferred to the psychiatry department.

During the several interviews with the patient and the family, it was discovered that the young man had dropped out of school and had not left the house for 5 years due to episodes of cyberbullying, living in self-imposed isolation and thus developing a hikikomori syndrome. He had also ceased regular eating. Known to social services, the mother and child's situation had been reported to the local psychiatry service years earlier. With the outbreak of Sars-CoV2 infection, in the two years prior the patient's admission to the ED, there had been no contact with the general practitioner, the psychiatric services, or other medical facilities. Due to the precarious social conditions, the patient was discharged into the care of mental health services after 24 days of hospitalization.

The patient overall health improved, as he still persistently receives nutritional and psychiatric rehabilitation. A definitive psychiatric diagnosis for the patient is still to be determined.

Discussion

Anemia, defined as an Hb level lower than 12.0 g/dL in women and 13.0 g/dL in men,¹⁰ considering all the variety of underlying causes, is one of the most common conditions worldwide, with an approximate global prevalence of 23.1%.¹¹

Its significant burden makes monitoring essential for providing proper care, both at the population and individual levels. However, the sheer number of affected individuals represents a challenge for healthcare providers, who are often unable to adequately address both initial diagnosis and follow-up.

The patient in our case suffered from a severe malnutrition, responsible for its life-incompatible anemia, functional bowel intussusception and multiorgan failure with metabolic acidosis.

The focus of the acute resuscitation was focused on maintenance of peripheral perfusion, restoring Hb level and starting an adequate enteral nutrition.

The patient developed severe malnutrition as his diet primarily consisted of industrial crisps, processed bread, and pre-packed sliced ham, provided by his mother. In fact, the patient withdrew from social life and interactions since the age of 16, after being the target of some episodes of cyberbullying. This patient suffered from a psychiatric isolative disorder, that could be described as hikikomori. In this specific case, an undetermined psychiatric condition coexisted within the family, specifically in the mother-son relationship, leading to a severe domestic neglect and further estrangement from the rest of the family.

Even though not already listed in the Diagnostic and Statistical Manual of Mental Disorders 5th edition (DSM-5), the hikikomori syndrome is widely described in scientific literature. Hikikomori is a Japanese definition of a self-isolation condition, occurring mostly in young teenager to young adult men;¹² its prevalence in the general population sets around 1.6%.¹³ As per the literature, Hikikomori is a form of pathological social withdrawal or social isolation whose essential feature is physical isolation in one's home. The patient should meet the following criteria: i) marked

social isolation in one's home; ii) duration of continuous social isolation of at least 6 months; iii) significant functional impairment or distress associated with the social isolation.¹⁴

In general, hikikomori is considered a non-psychotic phenomenon, distinguishable from the state of self-enclosure based on positive or negative symptoms of schizophrenia. However, there are cases of hikikomori coexisting with a variety of psychiatric disorders such as anxiety disorder, obsessive-compulsive disorder, or personality disorder.² In the cohort affected by psychiatric conditions partially overlapping with hikikomori the prevalence is variably increased. At present, it is still unclear whether psychiatric disorders give rise to hikikomori as a symptom or if the hikikomori condition could stand as the origin cause of coexisting psychiatric disorders.¹⁵

Why should an emergency physician be aware of this?

This case demonstrates the severe consequences of an untreated case of hikikomori, an emerging psychiatric condition that could lead to extreme neglect of basic nutritional needs. This case exemplifies the necessity for emergency physicians to expand their awareness on the phenomenon and remain vigilant for the signs of severe psychosocial disorders, for early detection and multidisciplinary care are vital to prevent such life-threatening presentations.

Furthermore, this case underscores the challenges faced by the out-of-hospital healthcare system. In the post-pandemic phase, frail and vulnerable patients struggle to adequately access elective care, which is often delayed until the critical point which leads to the emergency department.¹⁶ This role of the ED, less pronounced in the pre-COVID era, is expression of the difficulty within the National Health Service in intercepting the needs of the weakest categories of population. Proactive communication and network integration between emergency, social and out-of-hospital services are essential, along with collection and production of data on the social role of the ED as a surveillance function.^{17,18}

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