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CASE REPORT

A coin crisis: a case report of pica with minireview

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Abstract

Background Pica is defined as a feeding and eating disorder where there is consumption of nonnutritive substances not consistent with cultural practices or social norms. Its aetiology is still unknown, as its prevalence and optimal treatment, which seem to vary with patients' characteristics and the specific behaviours involved.

Objectives The authors present a case report of pica treated with copper supplementation, with further diagnostic and treatment considerations.

Methods Clinical records and interviews were used. A review was conducted using PubMed database.

Results A 59-year-old patient, diagnosed with paranoid schizophrenia, presented with sideroblastic anaemia secondary to zinc-induced copper deficiency in the context of pica. These behaviours ceased with copper supplementation. After 2-year remission, the patient was discontinued on oral copper. One week after, pica behaviours reoccurred, with further remission with renewed copper treatment. Based on temporal relationship, we propose that there might be an association between copper supplementation and pica, not related to its plasma levels.

Conclusions As far as we know, this is the first case report in the literature with such a long-term follow-up of pica treatment. Our findings challenge the cause–effect relation between micronutrient deficiency and pica and prompts further research in the non-adaptive theories of this poorly understood clinical entity.

Keywords Pica · Feeding and eating disorders · Anaemia · Sideroblastic · Zinc · Copper · Schizophrenia

Introduction

First described by Hippocrates, the eating of inorganic substances has long been recorded. The medical term to describe it—pica—comes from the Latin word for magpie (*Pica pica*), a bird that by folklore incessantly gathers objects to satiate its curiosity [1]. Pica is defined as a Feeding and Eating disorder by the Diagnostic and Statistical Manual of Mental Disorders, 5th edition, (DSM-5) where there is a consumption of nonnutritive substances not consistent with cultural practices or social norms during at least 1 month [2]. Many terms have been coined to describe pica behaviours according to the material ingested.

Pica prevalence is difficult to assess. It is underreported and the methodology of data collection varies among studies [3]. Recent studies report a prevalence of 37.5% among Hispanic pregnant women living in United States and of 41.6% in an African migrant population in France [1, 4]. Its exact aetiology is still unknown. It was found to be comorbid with major mental disorders (psychosis, obsessive–compulsive disorder, developmental, and neurodegenerative disorders) and co-occurring with other medical conditions, such as pregnancy and nutrient deficiencies. Some propose an etiological role for these associations, but none has yet established a causal relationship [3, 5].

Overexposure to heavy metals or other toxic elements; digestive disorders, such as constipation, intestinal occlusion or perforation; and parasitic infections, such as *Ascaris lumbricoides* and *Toxocara* spp, are some of the common medical complications of this disorder [6, 7].

Its treatment is not standardized, varying with patients' characteristics and their specific pica behaviours.

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Educational and behaviour modification approaches are proposed [5]. Reports describe successful treatment with micronutrient supplementation, antidepressants and antipsychotics. However, treatment reports are scarce and the evidence for efficacy limited [3].

Hereby we present a case report of zinc-induced copper deficiency in the context of pica, where the behaviour relapse and remission were found to be temporally related to oral copper supplementation.

Case report

We present the case of D.M., a woman 59 years old. Divorced at the age of 53, she lived in a rural area with her 28-year-old daughter and two grand-daughters (4 and 12-year-old). She had completed primary education at the age of 15, with learning difficulties, and used to work as a goldsmith until the age of 37 when she received her invalidity allowance due to her psychiatric condition.

Her first contact with mental health services was at the age of 22, being diagnosed with paranoid schizophrenia 6 years later. Since then she maintained ambulatory follow-up by the community psychiatry team, with nine hospital admissions as a result of psychotic decompensations during a 30-year period. Due to irregular medication compliance, she was started on depot antipsychotic early in the course of illness, in 2013 being stable with oral lorazepam 2.5 mg three times a day and haloperidol decanoate 200 mg monthly. There were reports of occasional alcohol consumption, with no other substances of abuse such as tobacco, opioids, stimulants or hallucinogens. She had no relevant psychiatric family or personal medical history.

In April 2013, the patient was admitted in the Surgery Emergency Department of Santo António Hospital (Porto), after ingesting multiple metal objects. She was brought by her daughter and, on admission, denied remembering the ingestion of those objects. These objects were removed by a laparotomy, with no complications. Once recovered, she was transferred to our psychiatry inpatient department for further investigation. On admission she was calm complying with interviewers; her mood was anxious with incongruent and blunted affect. The speech was poor in content, mentioning persecutory and paranoid delusions related to verbal auditory hallucinations. She described hearing multiple voices (female and male) localized in her outer space that threatened her. No command auditory hallucinations or delusional ideation related to pica behaviours were found. She provided no explanatory information for the ingestion, seemingly occurring as impulsive phenomena. No obsessive-like thoughts or anxiety relief were mentioned. According to her daughter, these behaviours had begun 2 months earlier with no trigger or environmental association. During the hospitalization, her antipsychotic medication was optimized (oral haloperidol 2 mg three times a day added) and was discharged a month later, with the diagnosis of acute episode of schizophrenia comorbid with pica.

A year later, in September of 2014, she was again admitted to the Emergency Department due to new psychotic decompensation, with the worsening of verbal auditory hallucinations. She also presented with persecutory and paranoid delusions, with the same characteristics of the prior admission. During routine admission workup, hematologic alterations were detected—leukopenia with severe anaemia (Hb 5.1 g/dL, on Table 1), prompting haematology evaluation. She was diagnosed with hematologic alterations due to severe nutrient deficiencies, and started on iron and folic acid supplementation. At discharge she was psychopathologically stabilized, with the diagnosis of paranoid schizophrenia, pica and bicytopenia; and prescribed with olanzapine 5 mg twice a day, lorazepam 2.5 mg three times a day, haloperidol decanoate 200 mg monthly and folic acid 5 mg and ferrous sulfate 357 mg supplementation once daily. In spite of the treatment, there was only partial resolution of the hematologic, which prompted a myelogram study that revealed the presence of 42% ring sideroblasts. Further tests were refused, such as lower and upper endoscopic study (with biopsy), and in the next 10 months she was treated with further 8 units of blood.

The patient was again admitted to our inpatient unit in June 2015, due to worsening of psychotic symptoms and treatment refusal—not only psychiatric but also blood transfusion for her anaemia (Hb 6.8 g/dL, on Table 1). During admission assessment she presented with generalized oedema and abdominal pain consistent with acute abdomen, prompting her referral to the surgical emergency

Table 1 Changes in blood values of the patient with pica disorder

	April 2013	September 2014	June 2015	December 2015	May 2016	August 2017	Reference values
White blood cell count ($\times 10^9$/L)	7.58	1.23	0.95	4.64	6.05	5.61	4–11
Hemoglobin (g/dL)	13.4	5.1	6.8	10.8	11.9	12.5	12–15
Copper (µmol/L)	–	–	1.4	20.5	17.57	17.59	11–24

The patient presented leukopenia and anaemia secondary to zinc-induced copper deficiency, which resolved after copper supplementation

department. Imagiologic investigation, with X-ray and abdominal computed tomography scan, was made revealing a voluminous metallic bezoar filling the gastric cavity (Fig. 1). A new emergency laparotomy was performed, with the extraction of "dozens of metal objects such as: coins, keys, screws, nail cutters, lighters, spoons and rings. The total weight of the objects was 1.941 kg". This episode enabled further haematologic study, this time in search for heavy metal poisoning. Reduced copper levels and raised zinc values were found, confirming the diagnosis of a zinc-induced sideroblastic anaemia secondary to zinc intoxication. Copper supplementation was then initiated—copper sulfate 2 mg daily with resolution of the hematologic disturbances (on Table 1). Due to the frequent psychotic decompensations and the precarious socio-familiar context, with insufficient support in the community, she was proposed for institutionalization and was admitted to long-term care in our hospital.

After 2 years under copper supplementation, adequate metal plasma levels and no acute psychiatric symptoms, the patient was discharged from haematology care and discontinued on oral copper. A week after the supplement suspension, the inpatient nursing team reported the start of nightly wake-ups and wandering behaviours, searching for metal objects in the ward. As reoccurrence of pica behaviours was suspected, an X-ray was made and confirmed the renewed ingestion of coins and other everyday metal objects. No ward functioning, therapeutic changes or organic intercurrences were found associated with these events. After consultation with haematology assistant, oral copper was reintroduced, and these behaviours ceased during the first week of supplementation. Since then, no other pica behaviours or acute psychotic symptoms were reported.

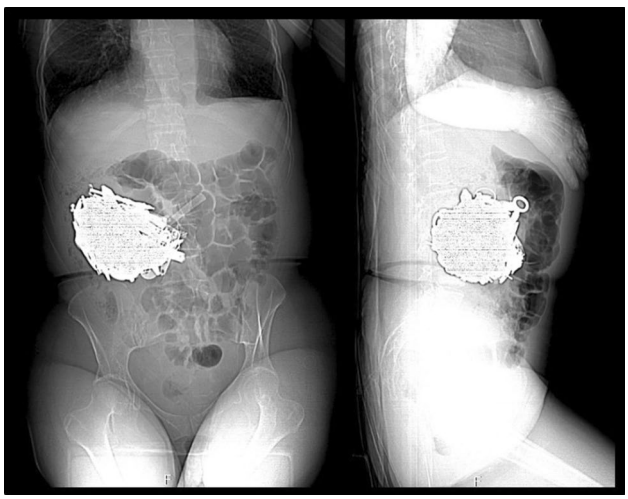

Fig. 1 Abdominal X-ray revealing a voluminous metallic bezoar filling the gastric cavity

Discussion

We present a case report of zinc-induced copper deficiency in the context of pica. Our patient ingested large amounts of metal. It was not possible to quantify the total amount of zinc and other metals ingested, but since a great number of euro and fifty cents coins—with 1500 mg and 390 mg of zinc, respectively—were removed from her stomach, we can assume that the daily ingestion was above the tolerable upper intake level of 40 mg/d [8], reaching toxic levels.

Our patient presented both iron deficiency, leukopenia and anaemia with no response to iron supplementation. We consider that these alterations could be multifactorial: malnutrition, alcohol consumption, and zinc intoxication secondary to pica. Zinc is known to interfere with intestinal copper absorption, inducing hematologic disturbances by disrupting heme synthesis. It induces intestinal synthesis of metallothionein, an intracellular ligand with high affinity for copper. Newly absorbed copper is stored in metallothionein which impedes its migration from the enterocyte to circulation [9]. Copper deficiency not only impairs iron absorption in the reticuloendothelial cells in the digestive tract, but also interferes with the action of mitochondrial enzyme cytochrome oxidase (a copper-dependent enzyme) responsible for heme synthesis. This frequently results in an iron-unresponsive anaemia, as reported in our case. Other causes of copper deficiency are described, mostly pertaining to diseases such as coeliac disease and inflammatory bowel disease. These typically present with other gastrointestinal symptoms, not found in our patient. Unfortunately, as the patient refused to perform lower and upper endoscopic study, we cannot rule them out entirely, despite being clinically unlikely. The mechanism of neutropenia in copper deficiency is also reported but is far less understood [9].

In our case, it is difficult to conclude what originates pica behaviours. There are in the literature different etiological theories to explain pica behaviours. First, cultural factors play a major role. For example, in some African populations geophagy is widespread and it is believed to reduce vomiting and to prevent infections. Indeed, the DSM-5 recognizes the importance of cultural background and alerts for the dangers to over-pathologize a practice that is viewed as beneficial in many populations [6]. Second, there are psychiatric comorbidity theories, namely psychotic disorders. Cases are described where patients with a priori mental illnesses present delusions and/or hallucinations that can originate pica behaviours, and where the treatment of those symptoms consequently resolves pica. This was not our case, since no relation to psychotic symptoms and pica behaviours was found. Third, some authors propose pica may be etiologically related to

nutrients' deficiencies, describing adaptative (such as the nutrient deficiency correction) or non-adaptative hypothesis [10]. In our case report, the blood tests of the first inpatient admission in April/2013 did not reveal hematologic disorders but no information exists on micronutrient status (this study is not routinely conducted). D.M. could have developed pica as a remedy for a nutrient deficiency not detected by routine exams. In favour of this hypothesis, is the fact that the pica behaviours ceased with copper supplementation. On the other hand, if it was just an adaptive mechanism, why do the behaviours return after normal blood levels where reached? Non-adaptative theories suggest pica behaviours to be an epiphenomenon of nutrient deficiencies that irreversibly alter neurological functions (e.g. appetite-regulating brain enzymes) or taste sensitivity causing non-food substances to become appealing [10]. In our case report, the pica behaviours ceased with copper supplementation on top of pre-existent neuroleptics, for more than 2 years, with recurrence of pica behaviours with the suspension of supplementation, despite normal blood levels. This treatment appeared to be effective both in acute treatment and in preventing relapses. Based on temporal relationship between the events and exclusion of other causes, we propose that there might be an association between copper supplementation and pica behaviours, not related to its plasma levels.

It is crucial for clinicians to be aware of pica, since, most of the times, patients omit these behaviours and the diagnosis is only established after etiological investigation of the associated complications. To prevent serious medical complications, pica patients should be assessed thoroughly, with routine physical examination, blood tests (with hemogram, renal, thyroid and hepatic functions, macro, and micronutrients status), stool test with microbiology analysis and an abdominal x-ray [5, 7]. Unfortunately, in our case report the initial blood test did not include macro and micronutrients evaluation, which could possibly have prevented further complications.

The literature is limited on case reports of pica and secondary zinc-induced copper deficiency. Although the treatment of its complications has already been described, the treatment of pica behaviours themselves has received less attention, and little or no information is given about long-term evolution and relapses. As far as we know, this is the first case report in the literature with such a long-term follow-up of pica treatment. Our findings challenge the cause–effect relation between micronutrient deficiency and pica behaviours and prompts further research in the non-adaptive theories of this poorly understood clinical entity.

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Compliance with ethical standards

Conflict of interest The authors report no conflicts of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional committee – Comissão de Ética do Hospital Magalhães Lemos, Porto with the form 87-C HML, and with the 1964 Helsinki declaration and its later amendments.

Informed consent Informed consent was obtained from the patient presented in this case report.

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