

CASE REPORT/CASO CLÍNICO

Pancytopenia Associated with Avoidant Restrictive Food Intake Disorder: A Case Report

Pancitopénia Associada a Perturbação de Ingestão Alimentar Evitante/ Restritiva: Um Caso Clínico

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Resumo

A perturbação de ingestão alimentar evitante/restritiva (ARFID) é uma das perturbações alimentares menos compreendidas, não havendo *guidelines* para o seu tratamento. Os doentes com ARFID frequentemente apresentam perturbações psiquiátricas comórbidas. As perturbações da alimentação e ingestão podem apresentar pancitopénia associada a um mau estado nutricional. O nosso objetivo é apresentar um caso clínico de pancitopénia num doente com ARFID. Trata-se de um homem de 20 anos internado por pancitopénia. Foi feito o diagnóstico de ARFID em comorbilidade com perturbação de ansiedade generalizada. Foi implementada terapia cognitivo comportamental, e após seis meses houve uma melhoria na ingestão alimentar, com maior diversidade de alimentos ingeridos. Este é o primeiro caso documentado de ARFID com pancitopénia. É crucial uma abordagem multidisciplinar, não só para um diagnóstico correto, mas também para o tratamento adequado com prevenção de recaídas.

Abstract

Avoidant restrictive food intake disorder (ARFID) is one of the least clearly understood feeding and eating disorders, and there are no guidelines for its treatment. Patients with ARFID often present comorbid psychiatric disorders. The feeding and eating disorders can present with pancytopenia associated with poor nutritional status. Our aim is to present a clinical case of pancytopenia in a patient with ARFID. The case concerns a 20-year-old man hospitalized with pancytopenia. It was diagnosed ARFID and comorbid generalized anxiety disorder. Cognitive behavior therapy was implemented, and after six months there was an improvement in the feeding behavior, with greater diversity of ingested food and remission of pancytopenia. This is the first documented case of ARFID with pancytopenia. A multidisciplinary approach is crucial not only for the correct diagnosis, but also for the effective treatment with relapse prevention.

Keywords: Avoidant Restrictive Food Intake Disorder; Pancytopenia

Palavras-chave: Pancitopenia; Perturbação de Ingestão Alimentar Evitante ou Restritiva

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INTRODUCTION

The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5),¹ includes avoidant restrictive food intake disorder (ARFID) as an eating or feeding disturbance manifested by a persistent failure to meet appropriate nutritional and/or energy needs, resulting in at least one of the following: significant weight loss, significant nutritional deficiency, dependence on nutritional supplements, and/or interference with psychosocial functioning.²⁻⁶ Based on clinical presentations, DSM-5 proposes three ARFID subgroups, including an apparent lack of interest in eating, sensitivity to the sensory characteristics of food, and fear of aversive consequences of eating.^{5,7} Children and adolescents within the first two ARFID subgroups are usually younger, include a greater proportion of boys^{4,7,8} and a longer duration of illness.⁸ ARFID in young children persist into later childhood, adolescence, and even adulthood.^{5,6,8} The etiology and management are poorly understood⁷ and there are no clear treatment guidelines.⁴ ARFID has a great likelihood of comorbid psychiatric illness, such as obsessive-compulsive disorder,⁹ anxiety disorders,^{3,4,9} depressive disorder,⁴ neurodevelopmental disorder.^{3,4,7,9} Eating disorders can present with pancytopenia,¹⁰ associated with serious vitamin deficiencies, mimicking hematological disorders. We report a case of pancytopenia in a patient with ARFID and relatively poor nutritional status.

CLINICAL CASE

A 20-year-old old single man was admitted to the emergency department in May 2019, with fatigue and malnutrition. He reported that he never ate any vegetable, meat, or fish, having refused to eat other food because he did not know the taste or texture. During 2019, he reduced the amount of food intake with the intention of losing weight, having lost 10 kg. Previously, his weight was 73 kg. There was no disturbance in body image or fear of gaining weight. On admission, the body mass index was 20.6 kg/m². Dietary assessment history revealed that at 4 years of age he only ate rice, fried potato chips, pasta, bread, milk and sometimes apple. At the age of 10, started to refuse milk and fruit. At the age of 15, was diagnosed with anemia, and was treated with iron and vitamin B12. Family described picky eating habits since childhood. He denied laxative use, diet pills or diuretics. There was a personal history of anxiety disorder, with psychotherapy from the age of 4 to 14. No other relevant personal or family history. Laboratory findings revealed pancytopenia with anemia and vitamin B12 deficiency (hemoglobin 5.3 g/dL; white blood cell count 2.600 cells/ μ L; thrombocytopenia 60.000 cells/ μ L; vitamin B12 undetectable). Bone marrow aspiration was normal. Acute leukemia and other oncologic diseases were excluded. He was hospitalized in the Medicine Department with necessity for blood transfusions and B12 vitamin and folic acid supplements. He exhibited emotional immaturity and significant difficulties managing interpersonal and environmental stressors throughout his hospital stay. He was observed by Liaison Psychiatry and diagnosed with ARFID and comorbid generalized anxiety

disorder, with extreme sensitivity to texture or taste of certain food and fear of aversive consequences of new eating habits but recognizing the consequences of malnutrition. During hospitalization, he started cognitive behavioral therapy (CBT), with reduction in anxiety regarding eating, and he was also referred to a nutritional therapist, but he could not change the feeding pattern. Despite this, given that the blood findings slightly improved, the final diagnosis was pancytopenia secondary to malnutrition. After discharge, he maintained oral vitamin supplementation and CBT, with decrease fear of trying unfamiliar foods. It was not prescribed psychopharmacological treatment. He accepted a structured nutritional plan with gradual exposure-based therapy (he tried a “new” food every 2 weeks). Gradually he started to eat other foods, and six months later he ate some meat (chicken and pork), eggs, fruits (apple and orange) and some vegetables (carrot and cabbage). His complete blood count was normal at this stage, with pancytopenia remission and was discharged from the hematology consultation.

DISCUSSION

The nutritional consequences of ARFID remain poorly described. Some papers reported weight loss,^{2,4,8,11} microcytic anemia³ and one describes a case of subacute combined degeneration of the spinal cord due to multiple vitamin deficiencies.¹² Although there is a case in the literature of anorexia nervosa with pancytopenia,¹⁰ there are no other case reports describing ARFID with pancytopenia secondary to malnutrition.

In this case report, pancytopenia was not a comorbidity, but a consequence of ARFID secondary to malnutrition. The food avoidance exhibited by the patient since childhood, with extreme sensitivity to texture or taste of certain food and fear of aversive consequences of new eating habits, may be driven partly by anxiety, and may function to relieve anxiety, thus negatively reinforcing the avoidant behaviour.⁹ This case is consistent with the prevalence of anxiety disorder among patients with a “selective eating” profile that persists since childhood into adulthood.^{13,14} Restrictive behaviour can induce specific deficiencies related to the nature of the excluded foods.³

Treatment for ARFID must take into account the extent of nutritional compromise, impact on weight, and interference with social and emotional development or functioning, with associated distress or impairment.⁸ Eating disorders are accompanied by anxiety, therefore it seems plausible to treat ARFID with similar techniques as used for anxiety disorders.^{5,15} Recently, case reports/series have suggested promising approaches for older children, adolescents and adults with ARFID, using either family-based treatment (FBT), cognitive behavioral therapy, or other novel approaches.¹¹

There are no psychopharmacological guidelines for ARFID. Benzodiazepines can be considered for anxious patients in the short term.⁴ In some severe cases, olanzapine³ or mirtazapine⁴ have been proposed. Selective Serotonin Reuptake Inhibitors are not a good option, because of their

potential side-effects, and could exacerbate the feeding issues.^{2,4}

In the case described, no psychopharmacological therapeutics were introduced because there was a decrease in anxiety with CBT and the patient accepted a diet plan with progressive introduction of food and pancytopenia remission.

In conclusion, this is the first documented case of ARFID with pancytopenia and an accurate differential diagnosis

of pancytopenia is essential for the most appropriate treatment.⁴ Like traditional eating disorders, ARFID presentation includes chronic restrictive eating behaviors resulting in malnutrition and serious medical complications.⁷ A multidisciplinary approach is required,⁴ not only for the correct diagnosis, but also for the effective treatment with relapse prevention, including medical monitoring, nutritional intervention, and psychological therapy.⁸

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References

1. American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders. Chicago: APA; 2013. doi:10.1176/appi.books.9780890425596
2. Spettigue W, Norris ML, Santos A, Obeid N. Treatment of children and adolescents with avoidant/restrictive food intake disorder: A case series examining the feasibility of family therapy and adjunctive treatments. *J Eat Disord.* 2018;6. doi:10.1186/s40337-018-0205-3
3. Feillet F, Bocquet A, Briend A, et al. Nutritional risks of ARFID (avoidant restrictive food intake disorders) and related behavior. *Arch Pediatr.* 2019;26:437-41. doi:10.1016/j.arcped.2019.08.005
4. Naviaux AF. Management of ARFID (Avoidant Restrictive Food Intake Disorder) in a 12-year-old on a Paediatric Ward in a General Hospital: Use of Mirtazapine, Partial Hospitalisation Model and Family Based Therapy. *Psychiatr Danub.* 2019;31:421-6. <http://www.ncbi.nlm.nih.gov/pubmed/31488765>.
5. Dumont E, Jansen A, Kroes D, de Haan E, Mulken S. A new cognitive behavior therapy for adolescents with avoidant/restrictive food intake disorder in a day treatment setting: A clinical case series. *Int J Eat Disord.* 2019;52:447-58. doi:10.1002/eat.23053
6. Sharp WG, Stubbs KH. Avoidant/restrictive food intake disorder: A diagnosis at the intersection of feeding and eating disorders necessitating subtype differentiation. *Int J Eat Disord.* 2019;52:398-401. doi:10.1002/eat.22987
7. Pitt PD, Middleman AB. A Focus on Behavior Management of Avoidant/Restrictive Food Intake Disorder (ARFID): A Case Series. *Clin Pediatr.* 2018;57:478-80. doi:10.1177/0009922817721158
8. Bryant-Waugh R. Avoidant restrictive food intake disorder: An illustrative case example. *Int J Eat Disord.* 2013;46:420-3. doi:10.1002/eat.22093
9. Kambanis PE, Kuhnle MC, Wons OB, Jo JH, Keshishian AC, Hauser K, et al. Prevalence and correlates of psychiatric comorbidities in children and adolescents with full and subthreshold avoidant/restrictive food intake disorder. *Int J Eat Disord.* 2020;53:256-65. doi:10.1002/eat.23191
10. Takeshima M, Ishikawa H, Kitadate A, Sasaki R, Kobayashi T, Nanjyo H, et al. Anorexia nervosa-associated pancytopenia mimicking idiopathic aplastic anemia: a case report. *BMC Psychiatry.* 2018;18:150. doi:10.1186/s12888-018-1743-6
11. Eckhardt S, Martell C, Duncombe Lowe K, Le Grange D, Ehrenreich-May J. An ARFID case report combining family-based treatment with the unified protocol for Transdiagnostic treatment of emotional disorders in children. *J Eat Disord.* 2019;7:34. doi:10.1186/s40337-019-0267-x
12. Chandran JJ, Anderson G, Kennedy A, Kohn M, Clarke S. Subacute combined degeneration of the spinal cord in an adolescent male with avoidant/restrictive food intake disorder: A clinical case report. *Int J Eat Disord.* 2015;48:1176-9. doi:10.1002/eat.22450
13. Zickgraf HF, Murray HB, Kratz HE, Franklin ME. Characteristics of outpatients diagnosed with the selective/neophobic presentation of avoidant/restrictive

- food intake disorder. *Int J Eat Disord.* 2019;52:367-77. doi:10.1002/eat.23013
14. Keel PK, Kennedy GA, Wick MR. Eating disorders in children: Is avoidant restrictive food intake disorder a feeding disorder or an eating disorder and what are the implications for treatment? *F1000Research.* 2018;7. doi:10.12688/f1000research.13110.1
 15. Zimmerman J, Fisher M. Avoidant/Restrictive Food Intake Disorder (ARFID). *Curr Probl Pediatr Adolesc Health Care.* 2017;47:95-103. doi:10.1016/j.cppeds.2017.02.005