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Case Report

Scurvy: Still Relevant or not? A Case of Scurvy in Siblings with Avoidant/Restrictive Food Intake Disorder

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Abstract

Scurvy is a disease caused by vitamin C deficiency, although it is rare in the modern world, it can occur in vulnerable populations: children and adults with eating disorders, residents of the Arctic who do not have their own fruits and vegetables, and people with low social status.

The goal is to use the example of this clinical case to acquaint doctors with the variable clinical manifestations of vitamin C deficiency in children, in order to increase vigilance and timely prescription of cheap and affordable drugs.

A clinical case of scurvy is described in a 4-year-old child with an autism spectrum disorder who is selective in food, with laboratory-confirmed vitamin C deficiency. The girl was hospitalized in the pediatric department of the Volyn Regional Territorial Medical Association for the Protection of Motherhood and Childhood with complaints of acute lameness and a hemorrhagic rash on the skin, which had been bothering her for the past 6 months. During the past six months, she was treated on an outpatient basis - however, her condition persisted without improvement. Due to the rarity of the disease, the "diagnostic odyssey" was long. After numerous laboratory and instrumental examinations, the child was diagnosed with scurvy, and the appointment of therapeutic doses of vitamin C led to a complete recovery.

Conclusion: Although scurvy is rare in the modern world, it can develop in vulnerable populations. In children with acute lameness, it is important to collect a nutritional history. If the diet consists mainly of products poor in vitamin C, and the symptoms are typical for scurvy, it is worth examining the patient for the level of ascorbic acid in the blood.

Introduction

Scurvy is a disease caused by a deficiency of Ascorbic Acid (AA) in the body. Scurvy has been known for a long time, in particular, as the main cause of death of sailors and polar explorers. The AA was discovered by Albert St. Gyordi 100 years ago, for which he was awarded the Nobel Prize. This discovery put an end to scurvy, saving thousands of lives [1]. In 1747, James Lind, surgeon of the British Royal Navy, conducted an experiment on the board of his ship. Knowing the deadly consequences of scurvy, Lind selected 12 patients onto the board and divided them into 6 groups of 2 people. In addition to their daily rations, Lind provided one group with oranges and lemons, while the other groups received cider, vinegar, seawater, or a mixture of garlic, mustard, and horseradish. Those who received citrus fruits recovered quickly and completely, leading Lind to conclude that oranges and lemons prevented scurvy. Since then, citrus juices of oranges, lemons, and especially limes have been used by the Royal Navy to prevent and treat scurvy. James Lind published his findings in a Treatise on Scurvy in 1753 [2].

Research made in the 20th century showed that most plants and animals are able to synthesize their own vitamin C.

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However, scientists managed to identify that the human body is not able to do this due to the lack of the enzyme gulonolactone oxidase necessary for ascorbic acid formation. Ascorbic acid plays an important role in the hydroxylation of collagen. This important biochemical pathway allows each collagen fibril to form a homogeneous and flexible triple helix configuration in the connective tissues of our body [3]. Certain tissues, such as skin, gums, mucous membranes, and bones, contain a higher concentration of collagen and are therefore more susceptible to AA deficiency [4]. AA is also necessary for the synthesis of dopamine, norepinephrine, adrenaline, and carnitine. In spite of all these specific and unique functions, most people know vitamin C for its antioxidant properties and ability to improve iron absorption from the intestinal tract [5].

The human body needs a daily intake of vitamin C [6]. Since humans are unable to synthesize AA, we get our daily dose from food such as citrus fruits (oranges, lemons, limes, grapefruit) and vegetables (potatoes, tomatoes, broccoli, spinach, Brussels sprouts, red peppers). AA is thermolabile, so it can be destroyed during the heat treatment of products containing it. Vitamin C is absorbed from the intestinal tract and has a biological half-life of approximately 30 minutes. The body does not have a depot of AA, but it is known that some tissues have higher concentrations of the vitamin (leukocytes, adrenal glands, pituitary gland). Current recommendations for daily vitamin C intake are 90 mg/day for men, 75 mg/day for women and 15-45 mg/day for children. Patients with chronic diseases such as cancer or diabetes, or cigarette smokers, require higher doses in their usual diet [5].

In Ukraine, vitamin C deficiency is a rare phenomenon, since our traditional diet contains foods with a high content of vitamin C (potatoes, citrus fruits, tomatoes, red pepper). For newborns, the necessary amount of vitamin C comes with breast milk. The most common risk factors for the development of ascorbic acid deficiency described in the literature are as follows: alcoholism, abuse of narcotic and psychotropic substances, eating disorders (especially anorexia nervosa), food allergies, inflammatory bowel diseases, celiac disease, terminal chronic kidney disease (hemodialysis). In pediatric practice, scurvy most often develops in children with autism who have eating disorders and are selective in food [7].

Clinical manifestations of scurvy can appear after 2-3 months of insufficient use of vitamin C. The early manifestations of the disease are nonspecific (general malaise, lethargy, and increased fatigue), but with progression following typical signs appear: pronounced skin syndrome (follicular hyperkeratosis, hemorrhagic rashes from petechiae to ecchymoses), joint and bone pain, poor wound healing, gingivitis, tooth loss and caries, anemia. Clearly expressed musculoskeletal pain in children with scurvy can be caused by hemorrhage into the muscles or periosteum, which will be clinically manifested by lameness. Therefore, one of the reasons for the development of acute lameness in children with eating disorders can be scurvy. In a systematic literature review, among 86 children with scurvy, 90% had complaints of limping. Other manifestations of vitamin C deficiency were follicular hyperkeratosis of the skin, petechiae, gingivitis, anemia, and long wound healing. Vitamin C deficiency was confirmed in the laboratory, and characteristic features of magnetic resonance imaging of bones and joints were sclerotic and transparent metaphyseal bands with periosteal reaction and adjacent soft tissue swelling [5].

To treat scurvy, a patient should take vitamin C for several months. While positive clinical symptoms can be observed already 24 hours after the start of therapy [5].

Therefore, in the cases (as described above) when patients have clinical manifestations of vitamin C deficiency, it is important to study the child's diet in detail when taking an anamnesis, and in the case of obvious eating disorders, remember about such disease as scurvy.

The goal is to use this clinical case to acquaint doctors with the variable clinical manifestations of vitamin C deficiency, and to increase vigilance and timely prescription of cheap and affordable drugs.

The purpose of the study is to increase the awareness of doctors regarding the clinical manifestations of scurvy and their timely diagnosis based on the described clinical case.

The research was carried out in accordance with the principles of the Declaration of Helsinki. Informed consent of parents was obtained for the study.

Clinical case

The patient was a 4-year-old girl, one of the twins, who was hospitalized in the pediatric department of the Volyn Regional Territorial Medical Association for the Protection of Motherhood and Childhood. She had complaints of progressive lameness, up to a complete refusal to walk independently, hemorrhagic rashes on the skin, and poor wound healing. The complaints have been bothering her for 6 months, gradually intensifying, so the girl was examined by a pediatrician, neurologist, orthopedic traumatologist, and rheumatologist.

The patient was in the VI pathological pregnancy (threat of premature birth at 30 weeks of gestation), III physiological birth at 36 weeks of gestation, from twins, where there is also a boy (has the same clinical manifestations, but less pronounced). The pregnancy proceeded with the threat of miscarriage at 30 weeks. Physiological birth at 36 weeks. She was born weighing 2600 g, and exclusively breastfed until the age of 1.5 months, after which she switched to artificial feeding. Physical development was age-appropriate: she held her head at 1 month, sat independently at 6 months, started to stand up with support at 8 months, and walked independently at 12 months. Psychological development does not correspond to age. She has a delay in speech development, an eating disorder (from 6 months of age), and other signs of autism spectrum disorder. A typical diet consists of wheat-based cereals, cookies, and one type of yogurt. Vaccinated according to the National Immunization Program. She had uncomplicated respiratory tract infections, including COVID-19 at the age of 3.

Since January, the mother began to notice a limp in the patient, which progressed, and after a few months, she

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could not walk independently. Over the past six months, she has been observed by a pediatric neurologist and an orthopedic traumatologist at her place of residence. Due to the deterioration of the patient's condition, she was referred for consultation with a pediatric rheumatologist at the VOTMO for the Protection of Motherhood and Childhood.

Physical examination showed the following: 14 kg weight, 100 cm height, pale gray skin color, follicular hyperkeratosis on the skin of the upper and lower extremities, isolated petechiae, small wounds after mosquito bites that do not heal for a long time, and bleed. She has no signs of gingivitis, her teeth are healthy. Auscultation of the heart and lungs was unchanged, palpation of the abdomen was normal, and the liver and spleen were not enlarged. She walks with support, limps on her right leg, muscle strength is reduced, and there is pain in the right knee joint during active and passive movements.

Differential diagnosis was performed with rheumatological, oncological, and infectious pathologies.

Laboratory tests revealed mild hypochromic anemia, increased ESR (53 mm/h), and decreased serum iron and vitamin B9. Platelet levels and coagulogram were normal. Cerebrospinal fluid analysis was normal, and CSF analysis by polymerase chain reaction for DNA of EBV, CMV, toxoplasma, HSV½, HHV6 type, and herpes type 6 was negative. HIV infection, tuberculosis, viral hepatitis B and C, and Lyme borreliosis were excluded (taking into account living in an endemic region). Bone marrow aspiration biopsy: excluded systemic hematological disease. SCT according to the oncoscreening program (26.07.24): did not reveal any pathology.

MRI of the lumbar spine, hip joints, proximal femurs, right knee joint, and right tibia (06.09.24): A diffuse hyperintense MR signal on PD fs and a hypointense signal on Tl were obtained from the bone marrow of the metaphyses of the tibia, fibula and femur with the presence of sclerotic lines (Harris lines) parallel to the growth zones and reactive subperiosteal edema, which is characteristic of scurvy. – The described changes in the bone marrow and subperiosteal may correspond to manifestations of nutritional deficiency (scurvy) (Figure 1). The diagnosis was confirmed by laboratory tests: the level of AA in the blood serum was < 0.50 mg/l with a normal range (N – 2-20 mg/l). The sick girl was cared for by a multidisciplinary team of specialists, including a child psychiatrist, psychologist, neurologist, infectious disease specialist, hematologist, dermatologist, and rheumatologist. Therefore, taking into account the data of the anamnesis, objective, laboratory, instrumental examination, and consultations of related specialists: absence of vitamin C-rich foods in the diet, progressive lameness, follicular keratosis of the skin, poor wound healing, the presence of specific bands on magnetic resonance imaging, anemia, low vitamin C levels in the blood, and after differential diagnosis with other diseases, the girl was diagnosed with avoidant/restrictive food intake disorder complicated by scurvy, moderate iron deficiency anemia, folic acid deficiency, and childhood autism.

The treatment consisted of intravenous administration of AA, according to UpToDate recommendations, 100 mg 3 times a day for a week, then 100 mg once a day orally in tablet form until complete recovery. Already 36 hours after the start of therapy, pain in the right lower limb disappeared during movements, and a month later the child began to walk independently.

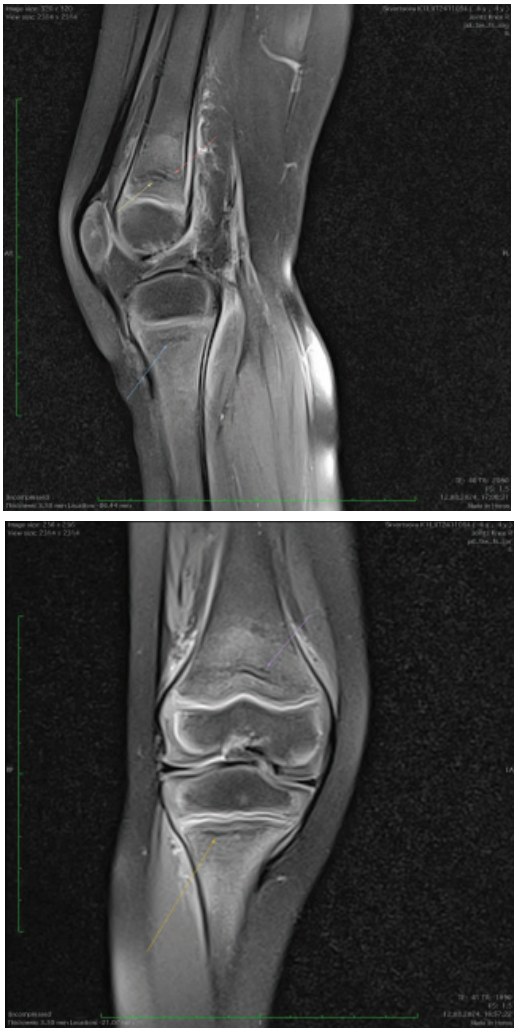

Figure 1: MRI of the knee joint. From the bone marrow of the metaphyses of the fibula, tibia and femur of the right lower limb, a diffuse hyperintense MR signal was obtained on PD fs and hypointense on Tl with the presence of sclerotic lines (Harris lines) parallel to growth zones and reactive subperiosteal edema.

Additionally, iron replacement therapy, folic acid, classes with a neuropsychologist, ABA therapy, classes with a speech therapist, fine motor skills ergotherapy, and constant observation by a child psychiatrist were prescribed.

After the appointment of therapeutic doses of vitamin C, positive dynamics were observed in the child after 36 hours: palpation and movements in the right lower limb were not painful, but she could not walk independently yet. After a month of taking vitamin C, she walks independently, still limps, petechiae are absent, residual phenomena on the skin after follicular hyperkeratosis. In parallel, therapeutic doses of vitamin C were prescribed to the boy twin, who also developed follicular hyperkeratosis, petechiae, and limping while the cause of these symptoms was being investigated in the girl twin. Their diet was the same: he ate the same foods as the girl. After 7 days of taking vitamin C, the symptoms disappeared.

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Discussion

This case was unique since almost all pathognomonic signs of scurvy were present in the girl at once. However, due to the rarity of the disease, difficulties in physical examination (the child is not in contact, and does not undergo examination), and collecting a nutritional history, scurvy was not suspected immediately. The clinical focus was more directed at the pain syndrome in the right lower limb and inflammatory changes in the blood. Differential diagnosis with rheumatological, oncohematological, and infectious diseases was carried out after many laboratory and instrumental tests. After a thorough examination of the patient's history, studying the girl's diet, a physical examination, and identifying specific signs on the limbs after MRI, she was suspected of having scurvy, and AA deficiency was laboratory confirmed. In this clinical case, the sick girl had all the typical symptoms of scurvy: skin, blood vessels, and bone tissue lesions [8,9]. In some of the described clinical cases, children also had gingivitis, fever, and tachycardia [10,11], while the sick patient did not have these symptoms. It is important to suspect ascorbic acid deficiency in time and start inexpensive treatment with vitamin C, without wasting resources on other examinations for patients with eating disorders, anorexia, severe depression, who do not have a balanced diet and have symptoms of scurvy: skin, bone and joint system and blood vessel lesions. And positive clinical symptoms will only confirm the diagnosis of scurvy a few days after taking vitamin C. The sick girl had positive dynamics after taking vitamin C on the 3rd day. Her condition improved, she was able to independently move her limb and stand on her feet. This clinical case shows the importance of carefully collecting a nutritional history in patients with eating disorders, depression, and anorexia in order to promptly suspect ascorbic acid deficiency and prevent mortality.

Conclusion

Scurvy is a rare disease in today's world, but it can develop in children with autism who have selective eating. Clinically, the disease is manifested by lameness, skin syndrome, gingivitis, and long wound healing.

In the case of children with autism spectrum disorders, pediatricians should routinely assess the patient's nutritional intake. If you suspect insufficient consumption of foods rich in vitamin C, you should determine the concentration of AA in the blood serum and prescribe it as a supplement to the daily diet.

For children with anemia, pain in the bones, joints, or muscles, as well as gingivitis and hemorrhagic rashes, who have eating disorders, scurvy should be included in the list of diseases for differential diagnosis.

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