CASE REPORT

Copper deficiency-induced bicytopenia caused by poor compliance in a paediatric patient with chronic malnutrition: A case report

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Abstract

A 12-year-old boy who underwent gastric wedge resection was transferred to our hospital because of vomiting, growth failure, and weight loss in January, 2016. We tried to restore his general condition by maintaining additional nutritional supply through peripheral parenteral nutrition (PN). However, continuous vomiting, weight loss, and superior mesenteric artery syndrome persisted because of low treatment compliance. The findings of hyponatraemia and bicytopenia did not improve. Bone marrow biopsy was performed, and it revealed copper deficiency. PN with additional micronutrient agents, including copper, were administered. In particular, invasive diagnosis and treatment, and adequate education improved the treatment compliance of the child. His copper deficiency and bicytopenia improved, and his weight and dietary intake also increased.

We confirmed that treatment compliance is important in paediatric patients with malnutrition. In chronic malnutrition, attention should also be paid to deficiency of micronutrients such as copper, which can lead to haematologic problems.

Keywords: Copper deficiency, Malnutrition, Intestinal failure, Adolescence

Introduction

Among children, those with intestinal failure (IF) is increasing in number because of short bowel syndrome, severe motility disorder, and congenital disease.[1] As chronic malnutrition can occur in paediatric patients with IF, nutritional support such as proper parenteral nutrition (PN) is highly important.[1] Complications or disorders after surgical treatment due to accident or bowel perforation are relatively common causes in older children without underlying disease.[2]

However, in the case of acquired bowel problem or accompanying chronic malnutrition, especially in adolescent patients, more careful treatment and various approaches, as well as medical treatment, are needed for general short bowel syndrome or IF.[3] The educational attainments of family members and relationships with other members of the family and society are also important considerations. If this treatment is not appropriate, psychogenic problems due to poor compliance may occur and secondary medical complications such as PN-associated liver disease or micronutrient deficiency may occur.[4]

We present the case of a patient with bicytopenia and persistent chronic malnutrition due to poor treatment compliance, who underwent surgical treatment with bowel perforation.

Case Presentation

A 12-year-old boy who underwent general surgery was brought to our hospital (Chung-Ang University Hospital,

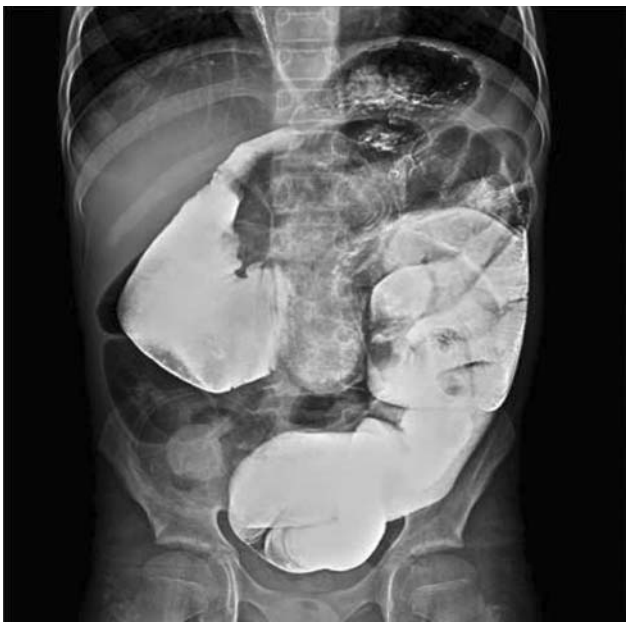

Figure-1: Gastrointestinal contrast enhancement findings confirming superior mesenteric artery syndrome with narrowed second and distal third portions of the duodenum. In the subduodenum region. Barium contrast remains in the small bowel and shows a transit delay pattern, indicating dilatation but no abnormalities such as obstruction.

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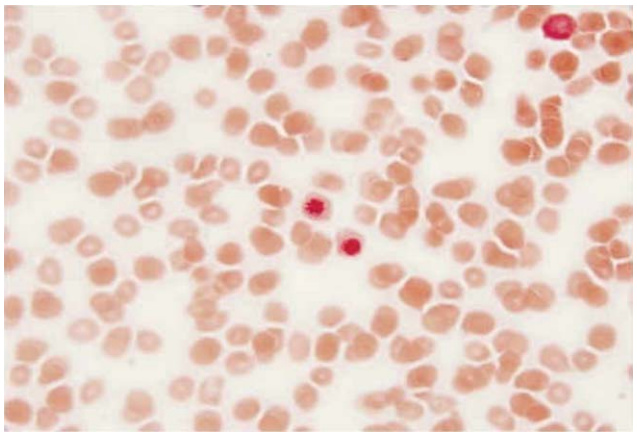

Figure-2: Bone marrow biopsy result showing several vacuoles in the granulocyte and erythrocyte global markers. Copper deficiency was suspected owing to an increase in histology. Cellularity is low, but no abnormality can be observed on iron staining, and no abnormal cells can be found.

Seoul, South Korea) in January 2016 because of chronic vomiting. The patient was subjected to an outpatient follow-up or chronic pseudo-obstruction and aerophagia at another tertiary medical institution about 3 years earlier. Two months later, the patient was subjected to exploratory laparotomy and gastric wedge resection for a gastric perforation. Ileostomy was also performed for subsequently occurring ileus and abdominal distension. The patient was admitted to our hospital because of persistent anorexia, weight loss, and low compliance, with ileostomy. Six months after the diagnosis of colonic pseudo-obstruction, total colectomy and ileostomy repair were performed. Thereafter, the repeated vomiting, anorexia, and weight loss persisted and acutely exacerbated.

At the time of transfer (January, 2016), his height was 138 cm (2.7percentile), body weight was 20.5 kg (<1 percentile), and body mass index was 10.76 kg/m^2 (<1 percentile). Two years earlier, his weight had increased up to 29 kg, and was 24.5 kg one month prior to hospital admission. At the time of presentation, he was in a chronic cachexic state, with the following laboratory values: sodium, 122 mEq/L; potassium, 3.3 mEq/L; chloride, 82 mEq/L; glucose, 88 mg/dL; serum albumin, 2.9 g/dL; total cholesterol, 86 mg/dL; triglyceride, 23 mg/dL. The basic complete blood count showed no signs of serious abnormalities as follows: haemoglobin, 11.1 g/dL haematocrit, 30.3%; white blood cell, 3,240/μL; absolute neutrophil count, 1,442/μL; and platelet, 313,000/μL.

As he was able to ingest orally, we recommended a high-calorie diet while encouraging oral ingestion, and limited his intake of beverages. We tried to restore his general condition and increase his weight by maintaining additional nutritional supply through initial peripheral PN. As he was eating a variety of foods, no medication other than total parenteral nutrition (TPN) and vitamin D was administered.

However, the continuous vomiting, anorexia, and weight loss persisted. Contrast imaging studies showed gastric emptying and small bowel transit delay, and superior mesenteric artery (SMA) syndrome. Abnormal findings such as obstruction were not confirmed (Figure-1). The patient was monitored through detailed interviews and by ward nurses. However, he kept going to other rooms every day at dawn and drinking a lot of carbonated drinks and juices. The weight loss worsened, and the SMA syndrome persisted. This was followed by continued parental education and self-education, and neuropsychiatry cooperation, but he did not show improvement. He often struggled with his nursing mothers and often showed a rebellious behaviour toward ward nurses.

The haematologic findings showed no significant abnormality related to the chronic malnutrition but showed bicytopenia, with decreased haemoglobin level of 7.5 g/dL, white blood cell count of 1,220/μL, absolute neutrophil count of 182/μL, and platelet count of 151,000/μL. His ceruloplasmin level decreased to 4 mg/dL. Decreased serum vitamin D and prealbumin levels, and osteoporosis were also detected in the imaging test. Bone marrow biopsy revealed bicytopenia due to copper deficiency (Figure-2). At that time, the copper level was 9 μg/dL (reference range, 75-145 μg/dL).

PN was increased by administering TPN via a central route in consideration of poor compliance. Additional micronutrient agents, including copper, were administered along with the basic TPN. This invasive diagnosis and treatment, and adequate education improved the treatment compliance of the child. His copper deficiency and bicytopenia improved, and weight and dietary intake also increased. The patient was also more satisfied with his weight gain and increased his amount of food intake. His relationship with surrounding people improved, and he also cooperated with the treatment. His body weight, which had decreased to 19 kg, increased up to 30 kg after treatment for 10 months. The results of the nutritional tests, including basic haematology and micronutrients such as copper, were also within the normal ranges.

Discussion

Copper is an essential micronutrient for haematopoietic

function, and copper deficiency is not a frequent occurrence in developed countries. However, in patients with chronic malnutrition and IF, copper deficiency and accompanying haematologic problems have been described in previous reports.[5-8] In a study of adults with copper deficiency, concurrent neurological and haematologic manifestations were more common and more than 50% of the subjects showed anaemia and leukopenia.[7] However, our present patient was of paediatric age and had no neurological manifestations. The possibilities of malnutrition due to absorption failure and micronutritional deficit, including severe copper deficiency, were low because of oral intake. The recurrent vomiting due to poor compliance and the resulting SMA syndrome may have been caused by the copper deficiency.

Currently, only medical treatments through PN or surgical treatments such as reconstruction or small bowel transplantation are considered as treatment options for malnutrition or IF.[3] However, for paediatric patients with IF, further treatment might need to be considered, especially in adolescent patients. A plan for growth such as height is also needed, and attention should be paid to the psychological state of the child. Careful mental care and management to build relationships with family members are also needed in situations such as long hospital stays.[9] This psychometric management is expected to improve long-term hospital life and compliance in adolescent patients who require long-term TPN.

Conclusion

The present case of malnutrition in a paediatric patient who needed long-term TPN, confirms that treatment compliance is important. In chronic malnutrition, attention should also be paid to the deficiency of micronutrients such as copper, which can lead to haematologic problems. In paediatric patients with IF, a multidisciplinary approach such as medical, surgical, and psychological management can prevent patient complications and ensure better life.

Ethics Approval and Consent to Participate: This study was conducted with the approval from the Institutional Review Board of the Chung-Ang University Hospital. Informed consent was confirmed by the IRB (1703-005-16049).

Consent for Publication: Written informed consent was obtained from the parent for the publication of this report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Availability of Data and Material: The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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