

Case Report

Auto-Brewery Syndrome in a Child With Short Gut Syndrome: Case Report and Review of the Literature

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Endogenous production of alcohol in the gastrointestinal tract of humans was first reported in 1948 (1). Several cases were reported particularly in Japan and then from around the world (1–8). The term “auto-brewery syndrome” has been used to describe patients who become repeatedly inebriated after ingestion of food of high carbohydrate nature in the presence of abnormal yeast proliferation, particularly of *Candida* species (8).

To our knowledge, the association of auto-brewery syndrome with short gut syndrome in children has not been reported. The purpose of this article is to describe one patient with this unusual syndrome and discuss its diagnosis and treatment.

CASE REPORT

A 13-year-old girl had long-standing short bowel syndrome secondary to jejunal atresia and necrotizing enterocolitis, and underwent extensive small bowel resection in the neonatal period. During the 6 months before her visit to this pediatric gastrointestinal clinic, she was noticed to have recurrent episodes of bizarre behavior, somnolence, disorientation, and a fruity odor of her breath and was suspected to be abusing alcohol. She was diagnosed as having alcohol intoxication when ethanol blood levels were repeatedly elevated, in the range of 250 mg/dL to 350 mg/dL. However, the patient persistently denied any intake of alcohol or alcoholic beverages. Nevertheless, this was suspected to be an adolescent behavior disorder. After repeated episodes of apparent intoxication, a psychiatric evaluation was performed and recommendation was made for admission for alcohol detoxification in a rehabilitation facility where she would be continuously monitored. While in the rehabilitation center, with no access to alcoholic beverages, she continued to have intoxicated behavior and again showed

elevated blood ethanol levels. D-Lactate levels were repeatedly undetectable. She was finally suspected to be producing ethanol endogenously by fermentation by a mechanism similar to D-lactic acidosis because of her short gut.

The family noted that these episodes tended to be observed frequently after ingestion of excess carbohydrates and juices. The father, who is a physician, had started to monitor her ethanol level using a commercial breath analyzer in relation to meals and continued to monitor the child for access to alcoholic beverages. A strong correlation was found between her elevated ethanol levels and intake of high carbohydrate meals or fructose-containing drinks. Her symptoms did not resolve with empiric courses of Bactrim® (Roche Pharmaceuticals, Nutley, NJ), Flagyl® (Pharmacia, Peapack, NJ), and Augmentin® (SmithKline Beecham, Philadelphia, PA) for suspected bacterial overgrowth. She then underwent upper gastrointestinal endoscopy study to obtain aspirates from her small intestinal fluids, which were sent for bacterial and fungal cultures. The aspirate grew abundant amounts of two types of yeast: *Candida glabrata* and *Sacchromyces cerevisiae*. After appropriate antibiotics were given based on fungal sensitivity studies (fluconazole), the symptoms resolved and there was no recurrence of the elevated ethanol levels.

DISCUSSION

This is the first case in the pediatric literature, to the best of our knowledge, to be reported linking endogenous ethanol production to short bowel syndrome and yeast overgrowth. An extensive review of the medical literature, using a standard computerized MEDLINE search, showed only a few reports about spontaneous endogenous alcohol production in humans and laboratory animals (1–4). In a report by Mezey et al., (2) a minimal production of postsurgical ethanol was noted in one third of patients after surgical jejunoileal bypass for morbid obesity. A similar occurrence was noted in a laboratory animal model. Mezey et al. (2) concluded that

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it is unlikely that ethanol production by bacteria in the intestine has a significant effect on the pathogenesis of liver disease after jejunoileal bypass. Ladkin and Davies (1) reported the death of a 5-year-old boy in Uganda who died as a result of the fermentation in his stomach of a large meal of sweet potato. The intragastric fermentation produced such large quantities of gas and alcohol that his stomach exploded, resulting in chemical peritonitis. His cultures grew mixed cocci and bacilli but no yeast. The Japanese patients described by Kaji et al. (3) had excess ethanol, which was produced by the fermentation that resulted from yeast of ingested carbohydrates. Three of these patients described were children aged 1 to 3 years, and none had short bowel syndrome. Infant formula fermentation with yeast was suspected to be a possible factor in production of ethanol by Bivin and Heinen (5) and a possible link to sudden infant death syndrome (SIDS) was suggested. However, this association was seriously disputed by Geertinger et al. (6) when they reported the rate of intestinal production of alcohol in vivo from *Candida*-dominated intestinal flora sufficient to exceed the normal alcohol metabolizing capacity of the liver. Therefore, measurable concentrations of alcohol in the blood from such cases would not be expected (6). Patients with D-lactic acidosis also exhibit altered behavior and neurologic symptoms, usually in relation to meals (7). However, acidosis should occur with elevated serum levels of D-lactate. The causative organism is usually a *Lactobacillus* species. None of these were present in the current patient.

The endogenous alcohol production, or auto-brewery syndrome (8), may probably be produced by a mechanism similar to D-lactic acidosis in short gut syndrome. An excess load of carbohydrates becomes available to an overgrowth of gut flora or yeast occurring in stagnant loops of a short bowel. The triggering role of excess carbohydrate load is also quite interesting; there may be

a need to modify the enteric feeding of patients with short gut syndrome to reduce the chances of such fermentation.

The current patient responded well to an antifungal agent, but this may prove to be an effective treatment but not a cure because of the development of resistant organisms. A goal of total eradication of alcohol-producing yeast in a patient with short bowel syndrome may prove to be elusive. Furthermore, antibiotic treatment aimed at bacterial overgrowth may only promote overgrowth of yeast species. Astute clinicians know to suspect D-lactic acidosis in patients with short bowels with symptoms such as those mentioned. It now seems that endogenous alcohol production must also be added to the differential diagnosis.

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