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CASE REPORT

Hepatic glycogen deposition in a patient with anorexia nervosa and persistently abnormal transaminase levels

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Summary Anorexia nervosa and other eating disorders characterized by calorie restriction have been associated with a variety of hepatic abnormalities. Fatty steatosis has been described in eating disorder patients. We report the rare finding of glycogen accumulation in the liver in a patient with anorexia nervosa, which to our knowledge is only the second such case reported in the literature. This case highlights the importance of monitoring for liver abnormalities in patients with restrictive eating disorders.

Introduction

Anorexia nervosa (AN) is an eating disorder characterized by restriction of caloric intake relative to caloric needs, leading to a significantly low body weight, with persistent behavior that hinders weight gain and an inability to recognize the seriousness of the low body weight [1]. The known gastroenterological complications of anorexia nervosa include esophagitis, delayed gastric emptying, acute gastric dilatation, constipation and hepatic dysfunction [2]. Etiology of hepatic dysfunction in anorexia nervosa patients has been attributed to a wide variety of causes however we present here a rare case of hepatic disease caused by glycogenosis, or glycogen accumulation within the liver.

Case report

A 26 year-old female presented to the Emergency Department (ED) of our institution with complaint of abdominal pain, nausea and constipation. Her history at that time was

Abbreviations: AN, anorexia nervosa; ED, emergency department; BMI, body mass index; ALT, alanine aminotransferase; AST, aspartate aminotransferase; CT, computed tomography; SSRI, selective serotonin reuptake inhibitor; PAS, periodic Acid-Schiff; PAS-D, periodic Acid-Schiff with diastase.

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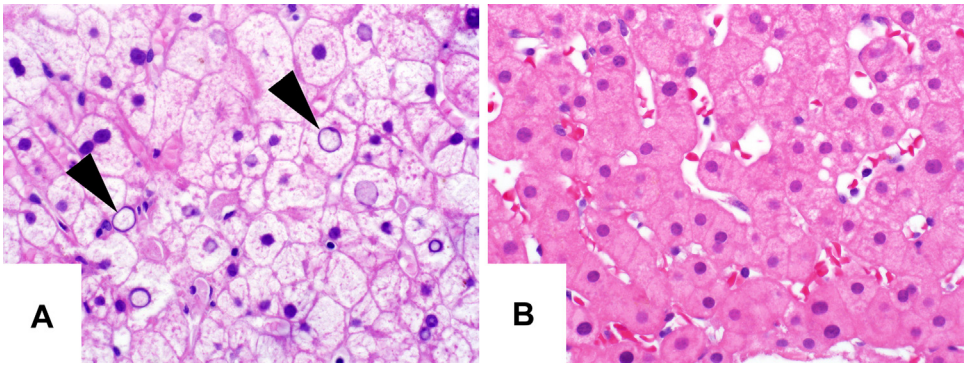

Figure 1 A. Hepatocytes with diffuse cytoplasmic clearing and swelling. Note the portal tract in the lower right with a minimal infiltrate and the absence of fibrosis (H&E, 200×). B. Normal hepatocytes.

remarkable for self-reported gluten intolerance, anemia of unclear etiology and secondary amenorrhea. She was not taking any medications; she had been on oral contraceptive pills but had discontinued them almost 1 year prior to presentation. She was notably underweight on exam, with a body mass index (BMI) of 16.5 kg/m^2. She was normotensive though bradycardic with a heart rate of 48 beats per minute. Laboratory evaluation performed during her ED evaluation was notable for hyponatremia with serum sodium of 130 mmol/L (reference range 135—145 mmol/L), alanine aminotransferase (ALT) of 101 IU/L (reference range 7—45 IU/L), aspartate aminotransferase (AST) of 79 IU/L (reference range 8—43 IU/L), and alkaline phosphatase 108 IU/L (reference rage 37—98 IU/L). Her complete blood counts, total bilirubin, serum amylase, lipase, serum creatinine and urinalysis were all within normal limits. Her pregnancy test was negative. Computed tomography (CT) of her abdomen and pelvis were negative, as were ultrasounds of her abdomen and pelvis, respectively. The patient was admitted to the Internal Medicine service in our hospital for further management of her abdominal pain. She was hospitalized for a total of 6 days, where all of her presenting symptoms as well as her abnormal laboratory values improved with fluid restriction, nutrition therapy and rest.

She established care with a primary care physician after that, who followed the patient closely in the year following. Her liver transaminases became persistently elevated again, to similar values as her ED presentation. She was referred to the Hepatology Clinic of our institution for evaluation. Testing undertaken to evaluate her persistently elevated liver enzymes included viral hepatitis serologies, anti-nuclear antibody, Celiac disease panel, smooth muscle antibody, anti-mitochondrial antibody, and anti-LKM1, and all were found to be negative. Liver biopsy was recommended, which the patient declined.

The patient eventually endorsed engaging in dietary restriction as well as bingeing and purging behavior for several years, accompanied by excessive exercise, anxiety, and distress around her eating patterns. The patient was diagnosed with anorexia nervosa. She was started on a selective serotonin reuptake inhibitor (SSRI) and enrolled in an outpatient eating disorder treatment program that included psychotherapy and nutritional rehabilitation. She subsequently gained weight and her eating disorder went into remission. She reported no further bingeing, purging, or food restriction.

Despite the reported improvement in her eating disorder for more than 6 months, the patient's ALT elevation was persistently three times the upper limit of normal and her AST was persistently two times the upper limit of normal. Because of this continued elevation, liver biopsy was recommended again. She finally agreed to undergo liver biopsy almost 24 months after her initial hospitalization.

The liver biopsy showed classic features of glycogenic hepatopathy [3]. The hepatic parenchyma showed an intact architecture without significant fibrosis and only minimal portal-based inflammation consisting of lymphocytes and macrophages. The dominant finding was in the lobules, where the hepatocytes showed diffuse swelling with cytoplasmic rarefaction (Fig. 1). The cytoplasmic swelling resulted in sinusoidal compression, making their identification difficult in some areas. Cytoplasmic membranes appeared distinct. Occasional nuclei also showed glycogenation. No significant steatosis, lobular inflammation, or hepatocyte necrosis was identified. A periodic Acid-Schiff (PAS) stain was positive within the hepatocyte cytoplasm. Following digestion with diastase, a PAS-D stain was negative, confirming the presence of glycogen in the cytoplasm (Fig. 2).

Unfortunately, following the liver biopsy she moved away to a different city and was subsequently lost to follow-up.

Discussion

This case describes the liver biopsy findings of glycogenosis in a patient with anorexia nervosa and elevated serum liver transaminases. The lifetime prevalence of anorexia nervosa in adults in the United States is estimated to be 0.6%, with a slightly higher prevalence of 0.9% in female adults [4]; these numbers are similar to prevalence rates in European cohorts as well. Unfortunately, the prognosis for patients with anorexia nervosa is poor, with high rates of psychiatric co-morbidities [5]. In one large review of anorexia nervosa patient outcomes, 46.9% of patients recovered completely, 33.5% had improvement in their symptoms though not complete resolution, 20.8% had chronically active disease and 5% ultimately died from either medical or psychiatric