

*Case Report*

# Thyrotoxicosis Masquerading as Superior Mesenteric Artery Syndrome in An Adolescent

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## Abstract:

Superior mesenteric artery (SMA) syndrome is a rare condition resulting in small bowel partial or complete obstruction. The third part of the duodenum becomes compressed between the aorta and the SMA once a subject loses weight rapidly. The patients at risk of SMA syndrome are those with eating disorders, trauma and substance abuse. We present a case of a 13 - year old female that complained of jaundice 3 months earlier, of recurrent attacks of vomiting and progressive weight loss. The vomiting was bilious and non-projectile following meals, 3-4 times per day. Upon presentation her weight was below the third percentile for age, with jaundice and hepatomegaly. Hepatitis proved to be autoimmune in nature and computerized tomography (CT) angiography confirmed SMA. The weight loss that preceded the SMA proved to be due to autoimmune thyroiditis and thyrotoxicosis. Duodenojejunostomy was not performed as she responded to carbimazole and nutritional support. Vomiting was controlled within a week. Hyperthyroidism can masquerade as SMA. Search for the underlying cause of weight loss leading to SMA is essential. We support the initial trial of conservative treatment for SMA.

## Level of Evidence of Study: IV (1).

**Keywords:** Superior mesenteric artery syndrome; thyrotoxicosis; autoimmune hepatitis; SMA.

**Abbreviations:** CT: computerized tomography; SMA: Superior mesenteric artery.

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## Introduction

The superior mesenteric artery (SMA) arises from the anterior aspect of the aorta at the level of the first lumbar vertebral body. It is enveloped in fatty and lymphatic tissue. SMA extends at an acute angle (between 38 and 65 degrees) with aorta into the mesentery (2). SMA syndrome arises in those with underlying inherent acute angle as little as 6 degrees when they rapidly/acutely lose the fat supporting the mesentery. The sagging of the duodenum results in compression of the third portion of the duodenum by the narrow space between the superior mesenteric artery and aorta (3). Affected patients with SMA syndrome present by bilious vomiting, weight loss, and the clinical picture of the cause. In patients with SMA syndrome, the primary cause of weight loss should be sought whether psychological or organic. Anorexia nervosa is notorious to cause complications including SMA syndrome, along with malignancies, and other debilitating conditions (4). SMA syndrome responds to conservative management in almost 60% of cases, and if not surgical duodenojejunostomy is indicated (5). We aimed to report that thyrotoxicosis may present as SMA syndrome, and discuss its association with hepatitis.

## Case Presentation

A 13 year old female, presented by jaundice, hepatomegaly, severe wasting, and bilious vomiting. Her parents reported that the condition started 3 months earlier by jaundice and paroxysms of bilious effortless non-projectile vomiting that was diagnosed as hepatitis virus infection. The jaundice resolved within 10 days, and paroxysmal vomiting worsened over time. Vomiting was bilious, and following meals, 3-4 times per day, approximately 2 cups, never blood tinged, and relieved by ondansetron.

The condition was associated with normal bowel habits and not associated with bleeding, fever, abdominal distention or swelling. She was feeding well with a normal appetite. However, she lost more than 5 kg in the past five weeks. The condition was associated with bone ache, tremors, palpitation and dyspnea on moderate exertion. There was no history of intentional dieting to achieve weight loss, skin rash, pigmentation, ulceration, bleeding following minor

trauma, fractures, easy bruising, joint affection, previous operation, previous blood transfusion, drug allergy, any other system affection or mental affection and the patient was performing well at school. The patient had congenital scoliosis and frequently wore a back-belt. By examination she was under-weight (26 kg, below 3rd percentile for age), her height was 148 cm (between 10<sup>th</sup> and 25<sup>th</sup> percentile for age), body mass index of 11.8 (below 3rd percentile for age, she was active, conscious, co-operative, oriented to time, place and person, with a senile facies and prominent zygoma with loss of cheek fat. The trachea was shifted to the right due to palpable diffuse thyroid swelling (5x3) cm which moved up and down with deglutition. There was subcutaneous fat loss from abdominal wall, limbs and buttocks. Apart from hepatomegaly, there was no other clinical abnormality. Her pubertal stage conformed with Tanner stage I, as she had no pubic hair, and underdeveloped breast buds. Fundus examination was normal. Psychological assessment was not conforming with eating disorder (not anorexia nervosa, bulimia nervosa, binge eating and eating disorder not otherwise specified, etc). Apart from the weight loss there was no system failure.

Investigations revealed that the hepatitis was type 1 autoimmune hepatitis, with positive antinuclear antibody (ANA) (titer more 1: 80) and positive anti-smooth muscle antibodies (SMAs) (titer more 1: 80) and no anti-liver kidney microsomal antibody type 1 (LKM-1). Primary hypothyroidism was evident with decreased thyroid stimulating hormone (TSH) of 0.01 mIU/L (normal range: 0.3-5 mIU/L), elevated free T<sub>3</sub> level of 12.7 pg/ml (normal range: 2-4.7 pg/ml) and T<sub>4</sub> level of 4.4 ng/dL (normal range: 0.7-1.8 ng/dL). Antibodies for thyroid peroxidase, thyroglobulin and anti-thyroid microsomal were positive, while thyroid stimulating immunoglobulin and thyroid receptor antibodies were negative. Hepatitis C virus and hepatitis B virus infections were ruled out. Coeliac disease, rheumatoid arthritis, and ulcerative colitis were also ruled out by negative serology and intestinal biopsy. Computerized tomography (CT) of abdomen and pelvis revealed severe gastric outlet obstruction and SMA syndrome with narrowed angle between the superior mesenteric artery and the aorta measuring 8 degrees. The child was diagnosed with autoimmune thyroiditis and autoimmune hepatitis with secondary SMA syndrome. She received carbimazole and nutritional support. Vomiting stopped within one week and the child was discharged without further need for surgical intervention to correct SMA syndrome. She received oral prednisone (with gastric protection using omeprazole) to control the autoimmune hepatitis. The enzymes normalized within 2 months. The girl had an uneventful follow up.

## Discussion

SMA syndrome is challenging in its diagnosis, and management. Index of suspicion is raised once bilious vomiting is present and other causes of intestinal obstruction are excluded. The CT angiography has greatly improved the certainty of diagnosis along with the typical endoscopic findings. The diagnosis of underlying metabolic cause of weight loss leading to SMA is very challenging, as intentional dieting and eating disorders are the primary suspects (4). Eating disorders are a diagnosis of exclusion and are described clearly in the American Psychiatric Association's Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5) (6).

Search for the organic cause of rapid weight loss resulting in loss of fat cushioning the SMA and development of SMA syndrome is essential, as treatment of the cause is an essential step in management. In this case report, the child suffered from primary thyrotoxicosis and autoimmune hepatitis that led to rapid weight loss within 3 month and SMA syndrome. It is very rare for thyrotoxicosis to present as SMA syndrome, with a few case reports (7). We are not aware of previous case reports in children with initial thyrotoxicosis presentation as SMA syndrome. Yet, the lesson learned here is that all causes of rapid weight loss should be ruled out. The remarkable rapid response to carbimazole, with complete resolution of vomiting was amazing and commands exclusion of thyroid disease in any child with SMA syndrome. The association of scoliosis and SMA was reported as a complication of scoliosis surgery (8), and as our case did not undergo scoliosis surgery. Hence scoliosis was ruled out as a possible cause for the SMA.

The other diagnostic challenge was hepatitis, which proved to be autoimmune type 1 hepatitis. Association between autoimmune hepatitis and thyroiditis has long been established. It is known that up to 45% of those with autoimmune hepatitis will develop thyroiditis (9). Other known associations are primary biliary cholangitis overlap syndrome as well. Other causes of hepatitis in this girl would have been coeliac disease, sclerosing cholangitis, ischemic, or viral infections (10) which were ruled out.

## Conclusions

Thyrotoxicosis may present as SMA syndrome, that might be associated with autoimmune hepatitis. Associations of autoimmune thyroid disease should be sought in children with thyrotoxicosis. Conservative management was effective in resolution of SMA syndrome in our child with SMA syndrome secondary to autoimmune thyroiditis and autoimmune hepatitis.

**Author Contributions:** Author searched medical literature, databases, conceptualized, conducted the case review and reviewed the final manuscript.

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## CONFLICT OF INTEREST

The authors declare no conflict of interest in connection with the reported study. Authors declare veracity of information.

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