

CASE REPORT**INABILITY TO SWALLOW TABLETS OFFERS AN OPPORTUNITY FOR DETECTION AND SUCCESSFUL TREATMENT FOR A RARE LIFETIME CAUSE OF DYSPHAGIA**Hailemichael Desalegn Mekonnen, MD, PhD^{1*}, Hari Conjeevaram, MD,²**ABSTRACT**

Introduction: This case presents an adult who was diagnosed with viral hepatitis C, and offering treatment for this potentially life-threatening illness provided an opportunity for diagnosis and therapy of a previously unrecognized long-standing cause of dysphagia.

Case Presentation: A 37-year-old female was diagnosed to have Hepatitis C virus (HCV) infection. Non-invasive assessment showed that she had moderate fibrosis and was recommended treatment with oral direct-acting antiviral (DAA) drugs. However, the patient was hesitant claiming that she had not been able to swallow any tablets for the last 30 years. Prior evaluation for her dysphagia was done with a barium swallow, which was reportedly normal. She was advised to have an upper endoscopy (EGD) evaluation, and it revealed a prominent proximal esophageal web. This was treated endoscopically and resulted in resolution of her long-standing dysphagia.

Conclusion: Causes of dysphagia can be obscure and lead to significant compromise in quality of life and inability to take life saving drug treatment. Esophageal webs can present with a long history of dysphagia and due to the proximal nature, could be missed by routine investigations. High index of suspicion is important to persuade for an endoscopy procedure in undiagnosed long-standing cause of dysphagia.

Keywords: Esophageal web; Dysphagia; Barium swallow; Upper GI Endoscopy

INTRODUCTION

This is a case of an adult patient with a long history of dysphagia whose diagnosis has been delayed due to a false reassurance by normal barium swallow study. The case was subsequently diagnosed as the patient was persuaded to undergo an upper gastrointestinal endoscopy (EGD) as she was unable to swallow tablet. The investigation revealed a proximal circumferential esophageal web.

CASE PRESENTATION

A 37-year-old female patient presented to our medical institution with a history of chronic fatigue and weight loss. The weight loss had been longstanding and she claimed that she has been basically consuming liquid diets and has trouble eating solid food including medications, for the last 30 years. The patient and her family believed the symptoms started since childhood, but the diagnosis was obscured for a long period. During her childhood, she has been evaluated at different centers, but to no avail, and the patients said that she started to 'live with it' being dependent on liquid and semisolid diets. General appearance showed a chronically ill-appearing woman with a BMI of 18kg/m².

On routine investigation, she was found to have a positive hepatitis C virus (HCV) antibody and subsequently was diagnosed to have ongoing viremia with a viral load of 582,000 copies/ml. Further investigations revealed that she has HCV genotype 4 infections and an abdominal ultrasound showed no obvious signs of cirrhosis. Other laboratory parameters showed hemoglobin of 11 gm/dl: all other laboratory evaluations (Liver chemistry tests, renal function tests, electrolytes) were unremarkable. HBsAg and HIV antibody test were negative. Fibroscan (Echosens 402) was done as non-invasive marker of assessing fibrosis severity and it was 9.0 suggesting moderate/severe fibrosis.

The patient was advised to undergo an upper GI endoscopy (EGD), which she strongly refused. For this reason, she was investigated with a barium swallow which showed a normal finding. This gave a false reassurance that her condition might be related to a globus sensation or some type of psychological stress. On the other hand, because of concern that she has significant hepatic fibrosis, it was recommended to treat with oral direct-acting antiviral (DAA) agents for her chronic hepatitis C.

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The patient, however, was very reluctant and declined therapy repeatedly claiming that she could not swallow any medication pills. She was finally persuaded to have an upper GI endoscopy with anesthesia support. The endoscope was difficult to pass down beyond the upper esophageal sphincter as there was a circumferential web, which was causing lumen narrowing and even the scope with a surrounding diameter of 8 mm could not pass [Figure 1]. This led to the diagnosis of a proximal esophageal web.

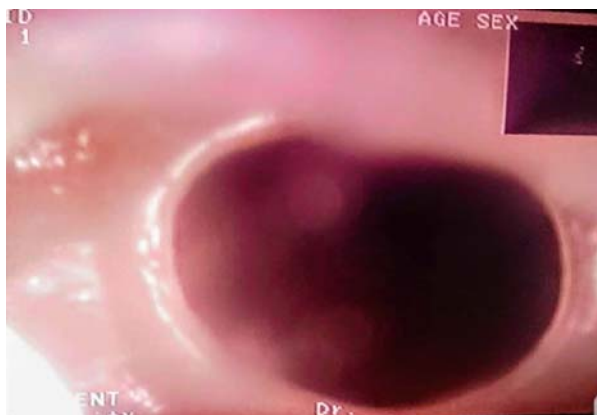


Figure 1: An upper esophageal web revealed on EGD in our 37-year old Ethiopian patient

The esophageal ring was then dilated with a pneumatic dilator cautiously to avoid aspiration and it was successful leading to rupture of the membranous ring (See Figure 2).



Figure 2: The image after an esophageal balloon dilatation has been done showing a rupture of the esophageal web

After the procedure, the patient was complaining of some discomfort while swallowing, which resolved slowly, but could not get herself to be convinced that she can swallow and be able to take her drugs. A subsequent pneumatic dilatation was performed after a week and at this time, the scope was able to pass and the remnant ring ruptured easily and the patient was given psychological support and reassurance. After this second therapy, the patient started eating solid food and she also started taking her drugs. She then showed marked improvement of her nutritional status. She successfully completed three months of DAA therapy (Velpatasvir + Sofosbuvir) and had a successful response with virological cure (sustained virological response/SVR) with undetectable virus at week 12 follow-up (SVR 12). She also slowly improved in her overall wellbeing with gratitude and has gained weight. It has been more than a year since she was put off follow up, and there are no complains of dysphagia.

DISCUSSION AND CONCLUSION

Esophageal web is an eccentric, thin < 2mm horizontal membrane and protrudes into the lumen of the esophagus. It is covered with stratified squamous epithelium and occurs anteriorly commonly in the cervical and middle esophagus. (1) It is associated with focal narrowing in the post-cricoid area and mainly affects middle-aged females (2). Patients are usually asymptomatic and the true prevalence is not clearly known. Patients may have dysphagia and regurgitation based on the site of the stenosis and especially when the stenosis is severe.

Hepatitis C Virus is one of the hepatotoxic viruses that could lead to chronic liver disease with liver failure, decompensation and liver cancer. It has affected 71 million of the world population and currently has effective therapies that can lead to cure within 2-3 months of treatment. (3) Hepatitis C has also an extrahepatic manifestations involving different organs of the body, but there was no reports of esophageal web in the literature. (4)

In patients undergoing barium esophagogram for dysphagia, 5 to 15 % of patients could have an esophageal web. In different literatures, it has been noted that patients usually do not become symptomatic until after 40 years of age (5). Our patient came to our attention at the age of 37 years, even though, she has had long standing intermittent symptoms. Her case could be explained as one of the postulated mechanisms, the developmental theory, where the webs are expected to occur when a pleat of mucosa is formed by in-folding of redundant esophageal mucosa due to shortening of the esophagus.

Esophageal webs are developmental anomalies with unknown pathogenesis. Unlike rings these anomalies rarely encircle the lumen but instead protrude from the anterior wall, extending laterally but not to the posterior wall (6). Webs are common in the cervical esophagus and are best demonstrated on an esophagogram with the lateral view. In up to 5% of cases, they are identified in an asymptomatic state, but when they are symptomatic they cause dysphagia for solids. Webs are fragile membranes and so respond well to esophageal bougienage with mercury weighted dilators (7). Symptomatic patients usually present with dysphagia to solids particularly hard ones (e.g., meat/bread), often intermittent, and patients may modify how they eat (chewing more thoroughly, etc.). In our patient, she had been avoiding swallowing hard solid foods including tablets and she was taking longer to chew before attempting to swallow foods.

Esophageal webs are associated with Zenker's diverticulum, dermatologic and immunological disorders, and iron deficiency anemia. There has also been a case report of celiac disease in association with esophageal web (8). In this current case, the patient's evaluation did not reveal any of the above associations or findings, although she was incidentally noted to have presence of chronic Hepatitis C Virus infection.

Literatures have shown that most children are asymptomatic, and, thus, they are far less detected and could adjust their life style and dietary habits as found in this case. This has been described as esophageal webs can present as a congenital manifestation and can stay until fourth decade without being diagnosed.

The diagnosis of esophageal web can be made by barium swallow study, however, if the esophagus is not distended adequately, it will give the appearance of subtle narrowing and can easily be missed (1). In a study published in 1985, the investigators found that barium detection rate can be evident in only 17 to 49 when proper techniques are not followed (9). Our patient had a normal barium swallow and this is an important lesson to improve our techniques in detection of esophageal webs and rings. It also showed that a normal barium swallow should not rule out upper esophageal pathology and additional investigations such as an upper endoscopy (EGD) may need to be done, especially in patients with long-standing symptoms. EGD shows a smooth, thin membrane that is usually non-circumferential and caution should be taken due to proximity to the upper esophageal sphincter. Once the diagnosis is made, management is usually performed with esophageal dilation and small webs usually rupture during the endoscope traverses the web. The goals of management are relief of dysphagia and the prevention of recurrent symptoms and patients usually have an excellent improvement on follow-up (10).

Our patient required sequential sessions of pneumatic dilations after which she had marked improvement of her symptoms and was able to complete taking her oral DAA drugs for her chronic hepatitis C infection resulting in successful treatment and achieving sustained virological response (virological cure). She was also able to have improved sense of well-being and she was able to eat more solid diets and also gained weight. In conclusion, we would like to state that patients with long standing dysphagia should be investigated for rare causes that can be easily treated with an upper GI endoscopy resulting in symptomatic relief and improved quality of life. In the case of our patient who was only 37 years old, unless an upper GI endoscopy had been done, the patient would have likely gone undiagnosed with persistent symptoms of her dysphagia and poor quality of life and potentially continued progression of her underlying chronic Hepatitis C virus infection.

Abbreviations: GI-Gastrointestinal, DAA-Direct Acting Antiviral, EGD- EsophagoGastroduodenoscopy, SVR-Sustained Virological Response, HCV-Hepatitis C Virus

Informed Consent:- The patient gave an informed consent about the publication;

PATIENT PERSPECTIVE :- Patient feels marked improvement from the intervention and happy that persuasion for the HCV treatment helped the diagnosis which will otherwise never be attempted.

Ethics approval and consent to Participate

Waiver request was secured from the Institutional Review Board at St. Paul's Hospital Millennium Medical College.

Consent for Publication: A written informed consent to publish this information was obtained from study participant.

Availability of Data and material:- The data is available from the corresponding author upon a reasonable request.

Competing Interest:-None

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Authors' Contributions

HD is the primary treating physician and responsible for writing the primary draft. HC has been a consultant in the case, reviewed the draft and both have agreed to submit the manuscript.

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