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*Case Report*

# An Unusual Presentation of Chest Pain and Laryngeal Discomfort in a Pregnant Woman: A Case Report and Literature Review

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**Intramural esophageal dissection (IED), characterized by bleeding into the submucosal space, leads to mucosal separation and dissection. The most prevalent symptoms are sudden chest or retrosternal pain, hematemesis, and dysphagia. Therefore, acute coronary syndrome and aortic dissection are among its most notable differential diagnoses. A 31-year-old pregnant woman presented with acute chest pain, laryngeal discomfort, and hematemesis. Emergency esophagogastroscopy revealed longitudinal mucosal dissection (upper esophagus to esophagogastric junction). The patient was successfully treated by avoiding the ingestion of solid foods. Clinicians should consider a diagnosis of IED for pregnant patients with acute chest pain, especially if hematemesis is present.**

**Key words:** chest pain, dysphagia, esophageal dissection, hematemesis

Intramural esophageal dissection (IED) is an uncommon condition resulting from bleeding into the submucosal layer with mucosal tearing [1]. IED typically manifests as sudden, intense retrosternal pain and at first may be mistaken for myocardial infarction or aortic dissection. The condition usually develops following endoscopy, or may occur in association with anticoagulation therapy or medical conditions with underlying bleeding diathesis. Adequate knowledge of the pathogenesis of IED may help clinicians avoid not only misdiagnosis but also unnecessary testing and treatment [2]. To the best of our knowledge, our patient is the first reported case of IED in a pregnant woman. In this case, the patient presented with acute chest pain, laryngeal discomfort, and vomiting of blood. Herein, we describe our experience in order to increase physicians' familiarity with this disease. We also provide a literature review on the etiology and pathogenesis of IED.

## Case Report

A 31-year-old healthy woman, 28 weeks pregnant, was transferred to our department with throat pain, chest pain, and laryngeal discomfort after repetitive episodes of hematemesis. The patient had an unremarkable medical history and was not on any medication. On physical examination, she was afebrile, alert, and hemodynamically stable with a heart rate of 78 bpm and a blood pressure of 112/70 mmHg. No oral or laryngeal injury was noted on visual inspection. Findings from her abdominal and cardiovascular examinations were unremarkable. Blood tests revealed a hemoglobin level of 11.3 g/dL, a normal coagulation profile, and normal liver and renal function. We sus-

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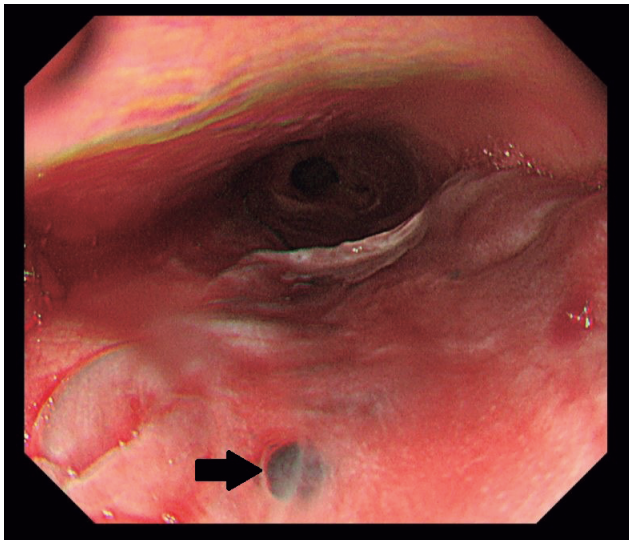

Fig. 1 The esophageal mucosa was longitudinally dissected from the upper esophagus to the lower esophagus, forming esophageal submucosal hematomas in some places (arrows).

pected upper gastrointestinal bleeding and performed emergency esophagogastroscopy. Endoscopy revealed longitudinal mucosal dissection from the upper esophagus to the esophagogastric junction. The submucosal bulge had dark blue discoloration and was non-pulsatile. The mucosa was friable without extrinsic compression of the esophageal wall (Fig. 1). Considering the patient's age, risk factors, symptoms, and characteristics of onset, we made a clinical diagnosis of IED. The patient was prescribed a soft diet for 14 days and was instructed to avoid solid foods. Her symptoms rapidly resolved, and she remained asymptomatic throughout her three-month follow-up.

## Discussion

IED was first reported in 1957 by Williams *et al*. [3]. The various names for this condition include "intramural rupture and intramural hematoma of the esophagus" [4,5] and "spontaneous submucosal hematoma" [6]. However, we recommend that the condition be called "intramural esophageal dissection" because this accurately describes the pathophysiology of the condition without implying a specific pathogenesis [7]. IED is defined as a longitudinal separation of the submucosa and esophageal muscular layers without perforation [8]. It is an intermediate esophageal injury ranking in severity between a superficial Mallory-Weiss tear and full-thickness Boerhaave syndrome. The most common signs are intense, sudden retrosternal pain (83% of patients), hematemesis (71%), odynophagia (41%), and dysphagia (32%) [7]. These symptoms are similar to those of other serious conditions such as pulmonary embolism, aortic disease, or intrathoracic esophageal disruption. Clinicians should suspect IED in patients who experience retching, vomiting, or forced swallowing of an impacted food bolus, and in those who develop dysphagia or hematemesis. A hematoma may restrict the esophageal lumen, resulting in dysphagia, and a dissected mucosa may perforate the esophageal lumen, resulting in hematemesis.

To the best of our knowledge, this is the first reported case of IED in a pregnant woman. IED can be associated with multiple etiologies and is most frequently seen in females and in patients on oral anticoagulants who suffer concomitant repeated vomiting. Sudden esophageal pressure changes resulting from uncoordinated movements like those induced by the Valsalva maneuver can cause this condition, as can complications of endoscopic procedures including eosinophilic esophagitis, nasogastric tube placement, or sclerotherapy [2,9]. In addition, rare etiologies of IED are associated with scenarios including severe eclampsia after delivery [10], extreme hyperextension of the back during yoga [11], and after cardiac resuscitation [12].

We searched PubMed for studies published between 1970 and 2022 associated with the terms "intramural (o)esophageal dissection" or "intramural (o)esophageal h(a)ematoma" and found 127 cases with available patient information written in English. The overall median patient age was 62 (44-75) years old, and the male and female patient median ages were 62 and 65 years old, respectively. The most common causes were idiopathic in 48 cases (37.8%), followed by dyscoagulopathy due to hemophilia or anticoagulation therapy in 40 cases (31.5%), and iatrogenic in 20 cases (15.7%). Regarding diagnostic tools, gastrointestinal endoscopy was performed in 96 cases (75.6%), and computed tomography (CT) and esophagography were conducted in 78 (61.4%) and 38 (29.9%) cases, respectively. Conservative treatment was selected in 98 cases (77.2%), while endoscopic procedures were performed in 18 cases (14.2%). Surgical intervention was required in 7 (5.5%) cases of refractory esophageal perforation. Although 119 patients (93.7%) were finally cured, 8

(6.3%) died due to exacerbation of comorbidities such as cirrhosis or coagulopathy.

Multiple modalities have been used to diagnose this condition. Contrast-enhanced CT is the preferred primary test as it is available in most centers and can be performed quickly. The typical appearance of IED on CT is a smooth, eccentric, intramural mass extending over a significant segment of the thoracic esophagus that may resemble a double barrel or dual lumen [13,14]. In cases in which CT is relatively contraindicated, as in our patient, IED can also be diagnosed endoscopically. IED typically manifests as a dark red to purple, smooth, non-pulsating, submucosal mass that compresses the lumen and bleeds easily on contact on endoscopy [15,16]. A raised, light-toned mucosa is seen on the surface, indicating that the hematoma is of submucosal origin. If the mucosal defect is of sufficient size, it may appear endoscopically as a "double-barrel esophagus." Considering our patient's pregnancy, the diagnosis was made endoscopically without radiological imaging.

The prognosis of IED patients is generally fair, as this condition is mild and can be managed without surgical treatment with several days of food abstinence and gastric secretion inhibitors.

In conclusion, we experienced a rare case of IED in a pregnant woman. Clinicians should consider IED in pregnant patients presenting with hematemesis and dysphagia-related chest pain. Gastrointestinal endoscopy is useful for diagnosis.

***Consent.*** Informed consent was obtained from the patient for publication of this case report and the accompanying images.

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