CASE REPORT

ESOPHAGEAL INTRAMURAL HEMATOMA

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Esophageal intramural hematoma is a rare form of acute esophageal injuries. We describe a case of esophageal intramural hematoma initially appearing as a longitudinal submucosal bulge at upper endoscopy. Thoracic computed tomography and esophagogram were performed for confirming the diagnosis. Endoscopic Biopsy was performed for the suspected esophageal tumor. The patient's symptoms resolved after biopsy and she resumed eating two days after admission. Endoscopy 7 days later showed a longitudinal esophageal ulcer and disappearance of the bulging mass. We suggest that early endoscopic intervention may improve life quality and shorten hospital stay.

Keywords: **intramural hematoma, esophagus**

Esophageal intramural hematoma (EIH) is a rare form of acute esophageal injuries, which include the more common Mallory-Weiss syndrome and Boerhaave's syndrome [1]. In contrast to Mallory-Weiss syndrome and Boerhaave's syndrome, EIH carries a better prognosis [2]. The disorder can occur spontaneously [3], or secondary to esophageal instrumentation [4-7], vomiting [8], trauma [9], pill-induced injury [10], food impaction related [11], and coagulation defects [12]. Treatment of EIH is usually conservative, and most patients have complete resolution after two to three weeks [7]. We describe a case of EIH, which appeared as a longitudinal submucosal bulge at upper endoscopy. Endoscopic biopsy for the suspected esophageal tumor with hematoma resulted in early relief of patient's symptoms and recovery of the hematoma.

CASE REPORT

A 63 year-old, previously healthy woman presented with acute onset of chest pain associated with dysphagia after a usual meal. The chest pain was dull, located over retrosternal area but no radiation to back or shoulder. There was no vomiting or hematemesis before the onset of pain. On presentation to our outpatient department, the patient was normotensive with systolic pressure of 120 mmHg. The rest of cardiorespiratory examination was normal. There was no evidence of melena. Electrocardiography revealed normal

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sinus rhythm with no evidence of ischemia. On a chest radiograph, the mediastinum was normal. Laboratory tests included a white blood cell count of 10000/ mm^3 (normal 4800-10800/ mm^3), a hematocrit of 38.1 % (37%-47%) and a platelet count of 381000/ mm^3 (130000-400000/ mm3). Prothrombin time was 10.6 seconds (normal 10-14 seconds), and activated partial thromboplastin time was 26.7 seconds (28-36 seconds). Carcinoembryonic antigen was 1.36 ng/ml (normal <5 ng/ml). Biochemical tests of cardiac enzymes as well as renal function were within reference ranges. Esophagogram revealed a sharply defined filling defect from mid to distal esophagus (Figure 1). Thoracic computed tomography (CT) with

Figure 1. Esophagogram revealing a sharply defined filling defect from mid to distal esophagus.

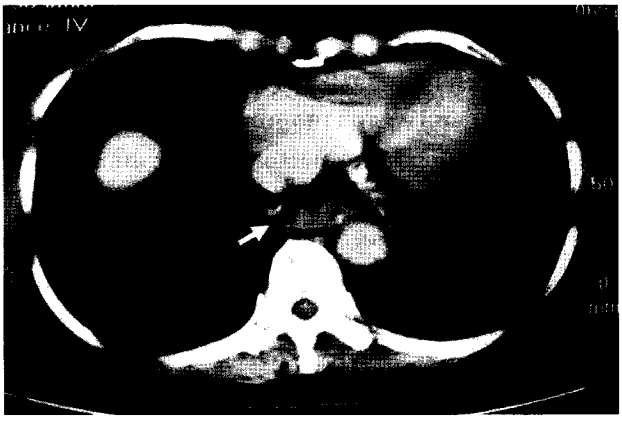

Figure 2. Thoracic CT with enhancement showing a water density mass without enhancement extending from carina to esophagogastric junction of esophagus (arrow)

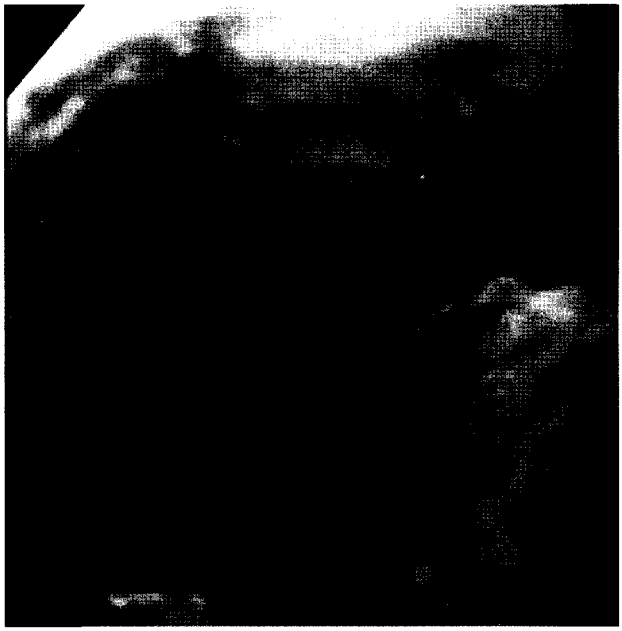

Figure 3. Endoscopic view of a longitudinal, fluctuant purplish submucosal bulge from mid to distal esophagus

enhancement disclosed a water density mass without enhancement extending from carina to esophagogastric junction of esophagus (Figure 2). The descending aorta was normal in appearance. Upper endoscopy showed a longitudinal, fluctuant purplish submucosal bulge from mid to distal esophagus (Figure 3). Because esophageal tumor could not be excluded, a biopsy specimen was obtained from the protruding lesion near esophagogastric junction. Histopathological assessment of the biopsy specimen revealed a picture of chronic esophagitis with marked congestion and hemorrhage. There was no evidence of atypical or malignant cells. A diagnosis of EIH was made. The patient was managed conservatively after admission with nothing by mouth, intravenous fluid, and a proton pump inhibitor. Her chest pain and dysphagia improved after the first endoscopy and she was free of symptoms on the second day. A soft diet was started on second admission day and increased to solid food on two days later. Endoscopy 7 days after admission showed a longitudinal ulcer from mid to distal esophagus and disappearance of the bulging mass (Figure 4). The patient was discharged on eighth hospital day. She followed an uneventful course and had remained well without sequelae after three months of follow up at out patient department.

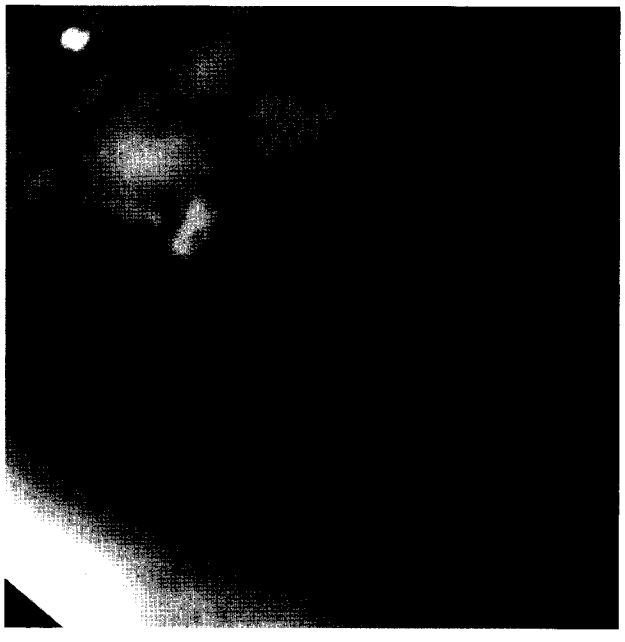

Figure 4. Endoscopic appearance 7 days later, showing a longitudinal ulcer from mid to distal esophagus.

DISCUSSION

EIH, also known as esophageal intramural rupture, esophageal submucosal hematoma, and esophageal apoplexy, was first described in 1968 by Marks and Keet [13]. It is generally thought that EIH is caused by sudden elevation of intraesophageal pressure, as occurs in Mallory-Weiss syndrome and Boerhaave's syndrome [1, 14], but the exact pathogenesis remains unclear. EIH usually presents with acute chest pain, typically accompanied by dysphagia, odynophagia, or hematemesis. Regardless of the etiology, the clinical triad of retrosternal pain, odynophagia or dysphagia, and hematemesis occur in approximately 35% patients. Nearly one-half of patients present with two of three symptoms [8]. In patients with normal hemostasis, EIH typically involves the distal esophagus. On the contrary, EIH tends to occur proximally in patients with impaired hemostasis [1, 8]. Unlike Boerhaave's syndrome and Mallory-Weiss syndrome, which occur commonly in men, EIH is found more frequently in women [15]. The mainstays of treatment have been nothing by mouth, intravenous hydration and hyperalimentation. Less than 15% of patients will ultimately require surgery [8].

The diagnosis of EIH relies on history, upper endoscopy, esophagogram, and contranst-enhanced CT. Endoscopically, EIH is typically described as a longitudinal blue-colored or purplish submucosal bulge [2]. Rarely, the lesion may mimic esophageal malignancy [16, 17]. Esophagogram may show a sharply defined filling defect or a sign of double-barreled esophagus [18], or a mucosal "strip sign" [8, 19]. Contrast-enhanced CT typically reveals a longitudinal mass of water

or blood density limited to the esophageal lumen without mediastinal bleeding [15]. The mass does not enhance with injection of contrast medium. Endoscopic ultrasound (EUS) can determine the extent of hematoma and is helpful in ruling out submucosal tumors [19]. If the lamina proporia is intact at EUS, the lesion can be termed a submucosal hematoma. When EIH is suspected, the differential diagnosis should include other emergent conditions, such as acute myocardial infarction, external compression of aortic aneurysm and aortoesophageal fistula. The most important disease to be differentiated is aortoesophageal fistula, which may appear as a bluish-gray submucosal mass. The clinical symptoms of aortoesophageal fistula typically demonstrate Chiari's triad of mid-thoracic chest pain, sentinel arterial hemorrhage, and exsanguinating hemorrhage after a symptom-free interval [20]. At endoscopy, aortoesophageal fistula is characterized by a pulsating tumor with an ulcer at the top, covered by adherent blood clots or with active hemorrhage [20, 21].

Suhamshu et al reported a case of EIH that was biopsied for ruling out an esophageal tumor [2]. Endoscopic biopsy resulted in early decompression of the hematoma and relief of symptoms three days after admission. Murata et al. also mentioned the usefulness of endoscopic intervention in a case report of EIH [22]. In the present case, a biopsy specimen was taken from the suspected esophageal tumor, also resulting in early improvement of symptoms. This might have been achieved by reduction of high pressure within the submucosal space and decompression of the hematoma. So, we suggest that early endoscopic intervention of EIH in patients with normal hemostasis may relieve patient's symptoms and improve life quality.

In conclusion, when facing patients with presentation of acute chest pain and dysphagia, EIH should be considered in the diagnosis. Differentiation of this disorder from acute myocardial infarction, aortic dissection, aortoesophageal fistula and esophageal tumor is important because of different treatment strategies and outcomes. Early endoscopic intervention may improve life quality and shorten hospital stays.

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食道壁內血腫

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食道壁內血腫（esophageal intramural hematoma）是較罕見的食道傷害。我們報告一個食道壁內血腫的病例，最初在上消化道內視鏡底下表現為一個縱走的黏膜下腫塊，胸部電腦斷層及食道鋇劑攝影確立了壁內血腫的診斷。由於懷疑食道腫瘤，我們做了切片檢查，病人在切片後即改善吞嚥困難的症狀，並在住院第二日即開始進食。住院第七日第二次上消化道內視鏡下所見只剩一個縱走的食道潰瘍。從這一病例報告，我們認為早期內視鏡治療可能會早期改善病人的症狀及縮短住院天數。

關鍵詞：壁內血腫、食道