
CASE REPORT

Spontaneous Oesophageal Intramural Haematoma: an Uncommon Condition in Two Patients

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ABSTRACT

Oesophageal intramural haematoma is an uncommon form of oesophageal injury. Oesophageal intramural haematoma can occur spontaneously without any preceding events. This report is of two patients who presented with haemoptysis and were incidentally found to have spontaneous oesophageal intramural haematoma by computed tomography. The diagnoses were confirmed by endoscopy and both patients were treated conservatively and had complete resolution confirmed by follow-up imaging and endoscopic examination. The typical clinical findings of central chest pain, dysphagia or odynophagia, and haematemesis, are non-specific and could mimic a wide variety of conditions. Correct diagnosis can be made with clinical history and characteristic computed tomography findings. Although spontaneous oesophageal intramural haematoma is uncommon and presentation with haemoptysis is not typical, radiologists and clinicians should not overlook this condition as a differential diagnosis.

Key Words: Esophagus; Hematoma; Hemoptysis; Tomography, X-ray computed

中文摘要

自發性食道壁內血腫：兩名患者的罕見病症

鄭加勁、柳洵

食道壁內血腫是食道損傷的一種罕見形式。自發性食道壁內血腫發病前可無任何先兆。本文報告兩名因咯血就診，於電腦斷層掃描中偶然發現自發性食道壁內血腫，後經內鏡確診的病例。兩名病人均接受保守治療，並於隨訪影像及內鏡檢查證實完全康復。該病的典型臨床表現如胸痛、吞嚥困難或吞嚥痛、及嘔血不僅均屬非特異性，而且類似多種其他疾病的症狀。不過結合臨床病史和CT特徵性表現可作出正確診斷。雖然自發性食道壁內血腫很少見，咯血也非典型起病症狀，但放射科醫生和臨床醫生不應忽視將該病列為鑑別診斷之一。

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INTRODUCTION

Oesophageal intramural haematoma (OIH) is an uncommon form of oesophageal injury associated with dissection of the mucosa from the muscular layer.^{1,2} OIH is part of the spectrum of oesophageal injuries that includes the more common Mallory-Weiss tear (mucosal tear) and Boerhaave's syndrome (transmural rupture).^{1,2} OIH can be classified into different categories according to pathogenesis: abnormal haemostasis, emetogenic, traumatic (including blunt trauma or food-induced trauma), iatrogenic, and related to aortic disease.² However, OIH can also occur spontaneously without any preceding event.³

Typical presenting symptoms are chest pain, haematemesis, and dysphagia or odynophagia.⁴ Diagnosis can be made radiologically, including computed tomography (CT), and endoscopically. Although OIH is well reported in the gastroenterology literature, little has been published regarding the radiological aspects.

This report is of two patients who initially presented with central chest pain and haemoptysis. OIH was identified by CT scan during the investigations for the clinical suspicion of pulmonary pathology, and the diagnosis of spontaneous OIH was subsequently established after endoscopy and clinical correlation. The patients were successfully treated conservatively, with complete resolution confirmed by follow-up imaging and endoscopic examination.

CASE REPORTS

Case 1

In November 2012, a 78-year-old man presented with central chest pain and massive haemoptysis. He had a history of hypertension and ST-elevated myocardial infarction, and was taking medication, including antiplatelet agent (acetylsalicylic acid 80 mg daily) and antihypertensive agents (metoprolol tartrate 75 mg twice daily and nifedipine sustained-release tablet 20 mg twice daily). He also had a history of Billroth II gastrectomy for gastrointestinal bleeding. On examination, he was haemodynamically stable. Cardiac, respiratory, and abdominal examinations were unremarkable. Chest radiograph revealed cardiomegaly without abnormal lung lesion. Electrocardiogram showed sinus rhythm and right bundle branch block. The initial haemoglobin level was 136 g/l (reference range, 130-170 g/l). Platelet count, international normalised ratio (INR), and prothrombin time were

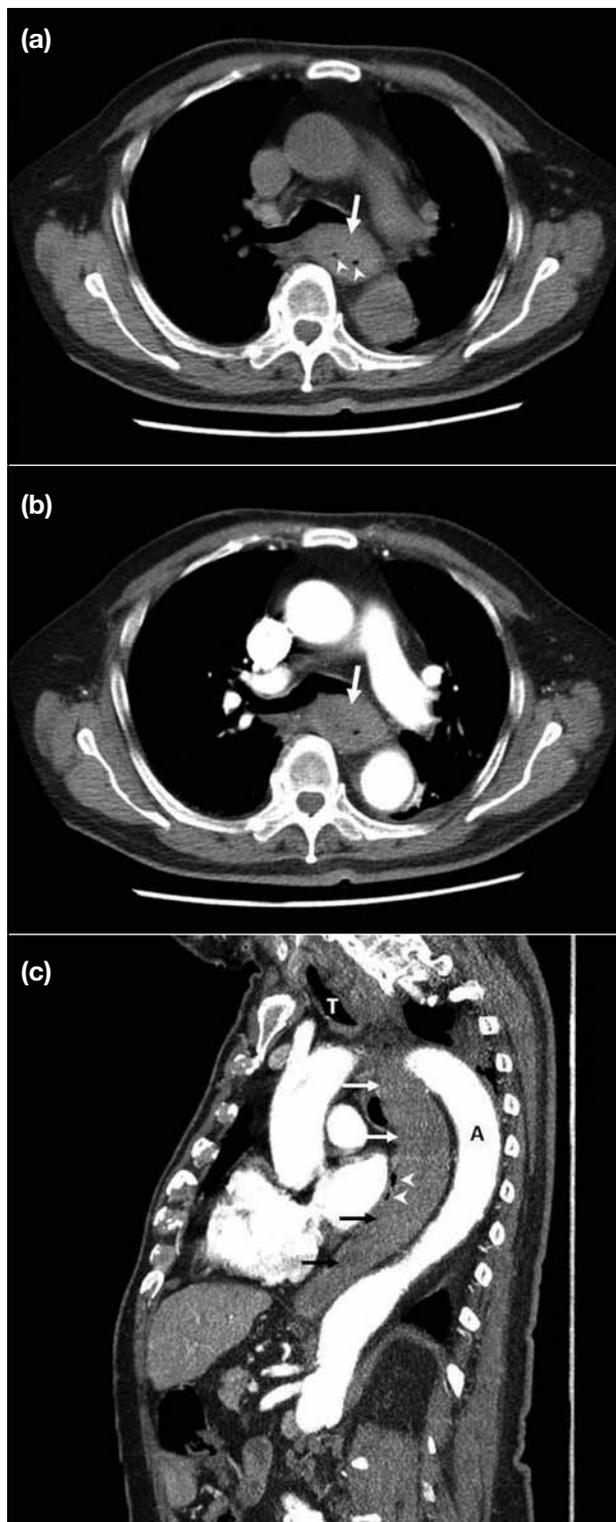


Figure 1. Computed tomography of thorax of case 1. (a) Unenhanced and (b) contrast-enhanced axial images at the level of the carina show homogeneously mildly hyperdense non-enhanced mural wall thickening (arrows) with markedly narrowed oesophageal lumen (arrowheads). (c) A contrast-enhanced sagittal reformatted image shows a long segment of eccentric intramural haematoma along the oesophagus (arrows) with markedly narrowed oesophageal lumen (arrowheads). Abbreviations: T = trachea; A = aorta.

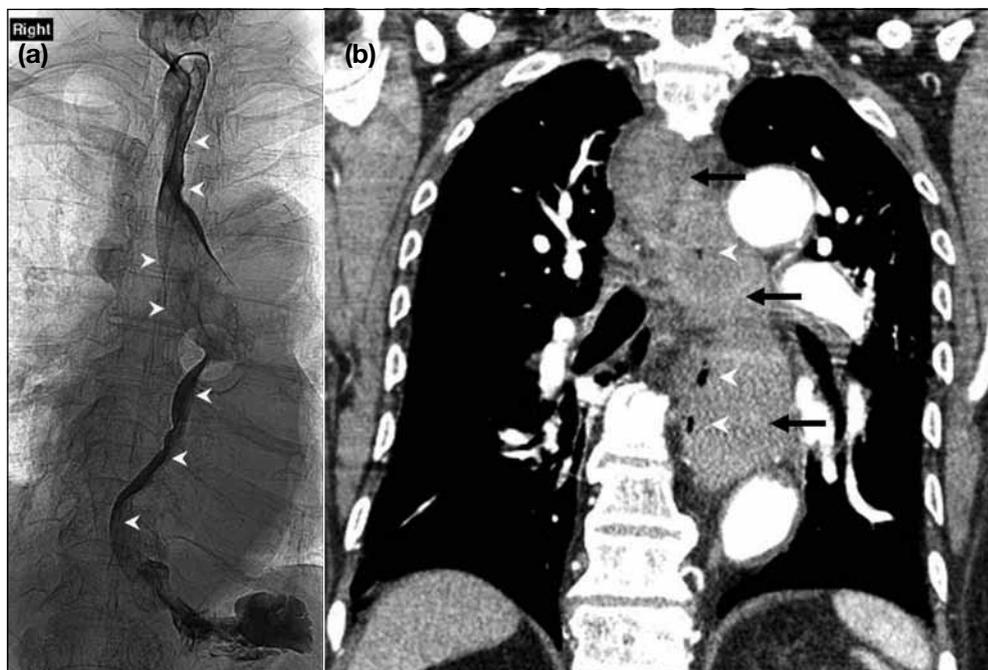


Figure 2. Water-soluble contrast swallow and computed tomography of thorax of case 1. (a) An image of the contrast swallow shows smooth luminal narrowing (arrowheads) along the oesophagus by long-filling defects. (b) A contrast-enhanced coronal reformatted computed tomography image shows eccentric intramural haematoma (arrows) with luminal narrowing (arrowheads), corresponding to the smooth-filling defects in (a).

normal.

Urgent CT bronchial angiogram was performed for planning of subsequent bronchial artery embolisation, which revealed neither focal lung lesion nor hypertrophied bronchial arteries. Instead, a long segment of homogeneously mildly hyperdense non-enhanced mural wall thickening with a markedly narrowed oesophageal lumen was detected (Figure 1). The findings were consistent with OIH. Supplementary water-soluble contrast swallow showed a long-segment of smooth-filling defects with luminal narrowing along the oesophagus, and reduced motility (Figure 2). No contrast extravasation could be identified.

Oesophagogastroduodenoscopy (OGD) performed 5 days after presentation confirmed intramural haematoma around 2 cm from the cricopharynx, extending all the way down to the distal oesophagus around 4 cm proximal to the gastro-oesophageal junction. No active bleeding was identified.

The patient was treated conservatively with nothing to eat or drink, intravenous fluid, proton-pump inhibitor (intravenous pantoprazole 40 mg every 24 hours), and antibiotics (intravenous amoxicillin and clavulanic acid 1.2 g every 8 hours).

On further inquiry, the patient had no prior history of emesis, blunt trauma, ingestion of foreign body, straining, or medical procedures involving the upper gastrointestinal tract before the symptoms arose. His symptoms gradually subsided after continuing conservative treatment. Repeated OGD 2 months after presentation showed resolution of the haematoma, which confirmed the results of the repeat CT scan performed 1 month after presentation (Figure 3).



Figure 3. Follow-up contrast-enhanced axial computed tomography of thorax of case 1 at the level of the carina confirms resolution of the oesophageal intramural haematoma (arrow).

Case 2

In May 2014, an 82-year-old woman presented with chest pain for 1 day. She had a history of hypertension, ischaemic heart disease, and cerebrovascular disease. The patient's medication included oral antiplatelet agent (aspirin enteric-coated tablet 100 mg daily) and antihypertensive agents (metoprolol tartrate 50 mg twice daily and nifedipine sustained-release tablet 20 mg twice daily). She developed shortness of breath and episodes of mild haemoptysis after admission. On examination, she was haemodynamically stable. Cardiac, respiratory, and abdominal examinations were normal. Oxygen saturation by pulse oximeter was 98% on oxygen supplement at 2 l/min. The initial haemoglobin level was 132 g/l (reference range, 130-

170 g/l). Platelet count, INR, and prothrombin time were normal. Chest radiograph was unremarkable.

The patient was suspected to have a pulmonary embolism, and subsequent CT pulmonary angiogram was performed, which showed homogeneously hyperdense non-enhanced density eccentrically located along the submucosal region of the distended oesophagus from the upper oesophagus down to the gastro-oesophageal junction (Figures 4a and 4b). Two disc-like hyperdensities, suggestive of tablets, were lodged in the markedly narrowed oesophageal lumen (Figure 4c). Findings were consistent with OIH. A small amount of extraluminal hyperdensity was also present around the lower oesophagus and descending thoracic

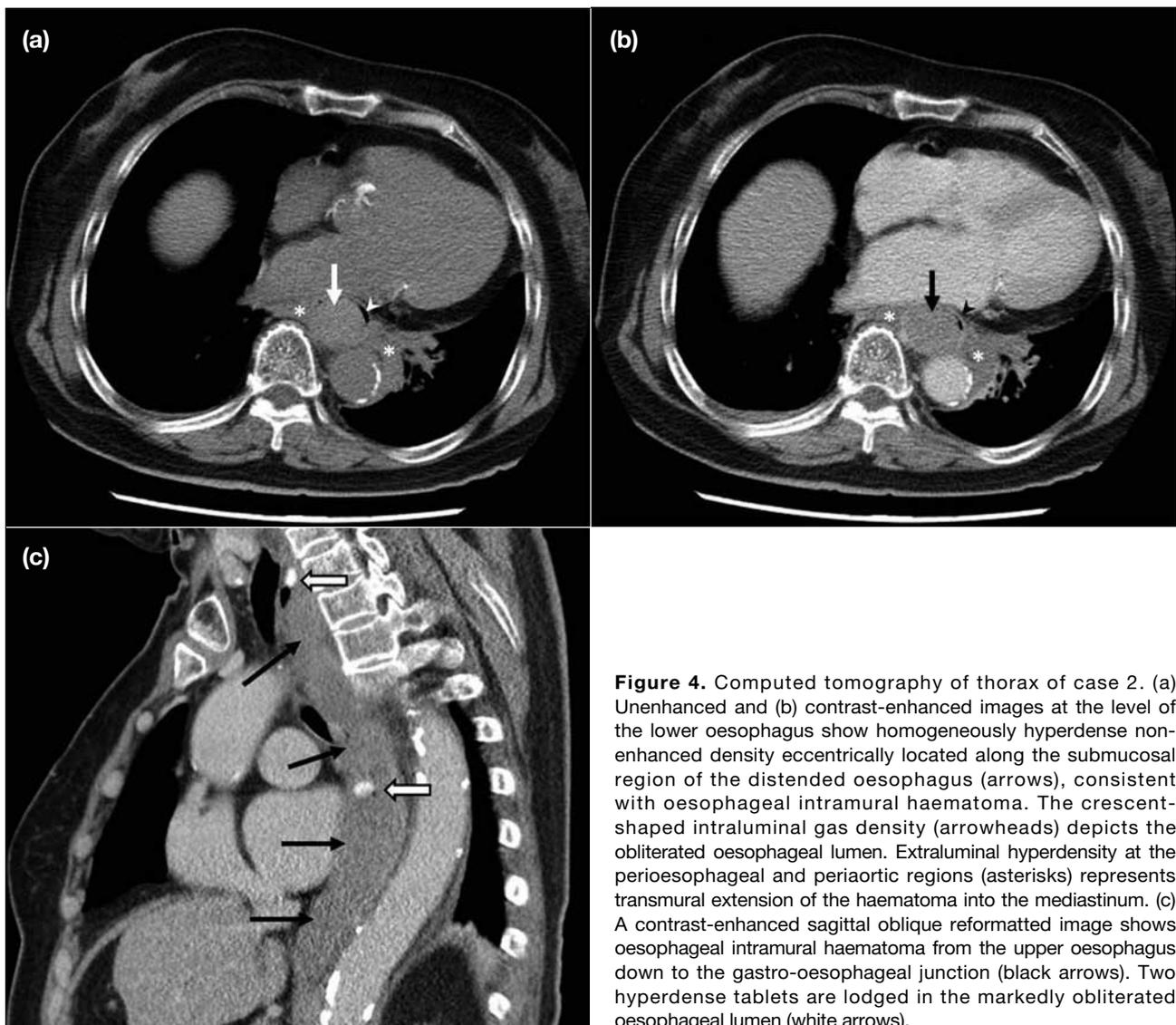


Figure 4. Computed tomography of thorax of case 2. (a) Unenhanced and (b) contrast-enhanced images at the level of the lower oesophagus show homogeneously hyperdense non-enhanced density eccentrically located along the submucosal region of the distended oesophagus (arrows), consistent with oesophageal intramural haematoma. The crescent-shaped intraluminal gas density (arrowheads) depicts the obliterated oesophageal lumen. Extraluminal hyperdensity at the periesophageal and periaortic regions (asterisks) represents transmural extension of the haematoma into the mediastinum. (c) A contrast-enhanced sagittal oblique reformatted image shows oesophageal intramural haematoma from the upper oesophagus down to the gastro-oesophageal junction (black arrows). Two hyperdense tablets are lodged in the markedly obliterated oesophageal lumen (white arrows).

aorta, which could represent transmural extension of the haematoma into the mediastinum (Figures 4a and 4b). There was neither pneumomediastinum nor pneumothorax to suggest perforation. The thoracic aorta appeared unremarkable.

Urgent OGD showed bluish intramural haematoma over the upper oesophagus 12 cm from the incisors, obstructing more than half of the lumen through which further advancement of the endoscope was not possible. No active bleeding was identified.

On further inquiry, the patient had no prior history of emesis, blunt trauma, ingestion of foreign body, straining, or medical procedures involving the upper gastrointestinal tract. The patient was treated conservatively with nothing to eat or drink, intravenous fluid, oral proton-pump inhibitor (pantoprazole 40 mg daily), and intravenous antibiotics (amoxicillin and clavulanic acid 1.2 g every 8 hours); and the symptoms gradually improved. The repeated CT scan 13 days later revealed resolution of the oesophageal haematoma (Figure 5). A barium swallow performed 18 days after presentation showed an unremarkable oesophagus apart from tertiary contractions, which were compatible with presbyoesophagus.

DISCUSSION

OIH is an uncommon clinical entity and is part of the spectrum of oesophageal injuries, which includes the

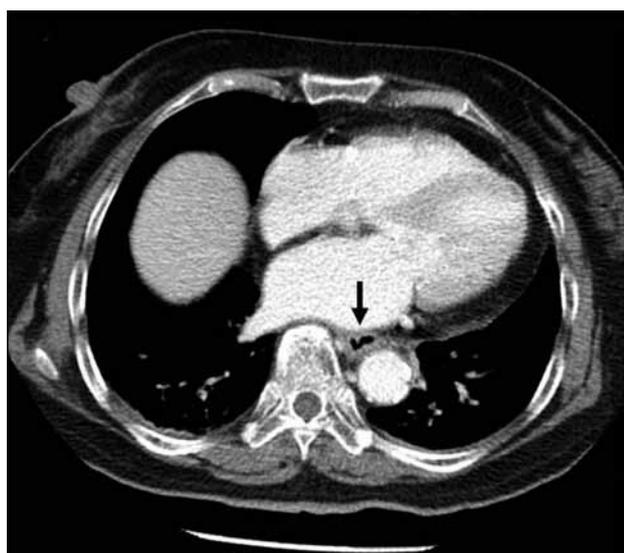


Figure 5. A follow-up contrast-enhanced axial computed tomography image of the thorax of patient 2 at the lower oesophageal level confirms complete resolution of the oesophageal intramural and mediastinal haematoma (arrow).

more common Mallory-Weiss tear and Boerhaave's syndrome.¹ OIH results from haemorrhage within the oesophageal wall involving the submucosal layer. Continuous submucosal bleeding could result in intramural dissection and even mucosal or muscularis disruption.²

Based on the pathogenesis and nature of the haemorrhage, OIH could be classified into different categories, including abnormal haemostasis, emetogenic, traumatic, spontaneous, and related to aortic disease. The term 'spontaneous' is used when OIH is not associated with vomiting, eating, trauma, abnormal haemostasis, or aortic pathology.⁵

Two of the three typical presenting symptoms of chest pain, haematemesis, and dysphagia or odynophagia are found to be present in 80% of the patients.⁴ Pain is usually gradual in onset, exacerbated by swallowing, and eventually becomes intense and severe, localised to the retrosternal or epigastric region. Haematemesis is, however, relatively infrequent but will eventually develop in 56% of patients as the haematoma ruptures through the mucosa.² To the best of our knowledge, there is no previous report of patients presenting with haemoptysis.

As the clinical findings are non-specific and could mimic a wide variety of acute conditions, including cardiovascular and pulmonary disease, further investigation is usually necessary. CT is a readily available, rapid, and non-invasive imaging modality that is useful for differentiating oesophageal pathology from other thoracic conditions.⁶

CT typically reveals concentric or eccentric oesophageal wall thickening with an intramural mass that is of blood attenuation, depending on the age of the haematoma. OIH characteristically shows no enhancement after intravenous contrast administration, except that secondary to aorto-oesophageal fistula. OIH involves a variable length of oesophagus and with variable degrees of oesophageal lumen obliteration. Air within the haematoma, but not in the mediastinum, may suggest mucosal tear or infection, while air within the mediastinum should be considered a sign of transmural rupture.² In case 2, haematoma was present at the perioesophageal and periaortic regions, which is suggestive of transmural extension of the OIH.

Contrast swallow examination should be done with

water-soluble contrast medium.⁷ Well-defined filling defects and narrowing of the oesophageal lumen with reduced oesophageal motility would be seen during the examination. Contrast extravasation could be demonstrated if there is communication between the haematoma and the oesophageal lumen due to mucosal rupture, producing a strip-like collection of contrast material inside the wall, namely the double barrel sign.^{8,9}

Oesophageal wall thickening per se is non-specific and may be seen in other conditions. Circumferential oesophageal wall thickening could be due to diffuse oesophageal spasm, oesophagitis or, occasionally, malignant oesophageal tumours.¹⁰ Differential diagnoses for eccentric mural thickening include malignant (primary or secondary) and benign neoplasms. Oesophageal wall thickening may also be mistaken for a distended oesophagus on plain chest radiography or CT in cases of achalasia.¹¹ Clinical history is helpful to differentiate different possibilities, as OIH presents with acute chest pain, haematemesis, and dysphagia or odynophagia, while progressive dysphagia and constitutional symptoms may point towards malignancy. Differentiation of OIH could be made by the characteristic blood attenuation and non-enhancement of intramural haematoma, in contrast to other conditions. The morphology and distribution may also give a clue to OIH.² If the diagnosis is in doubt, follow-up study could be performed to confirm interval resolution of the haematoma, ruling out the possibility of malignancy.^{12,13}

Spontaneous OIH generally has a benign course and treatment is mainly conservative with parenteral nutrition, acid-suppressing medication, and antibiotics.¹⁴ The haematoma should resolve in 1 to 3 weeks, with complete healing of mucosal tear and recovery of normal wall tone and peristalsis.¹⁴

CONCLUSION

This report is of two patients with spontaneous OIH that were incidentally detected by urgent CT. Both diagnoses were confirmed by endoscopic findings and complete resolution on follow-up CT. Conservative management was efficacious for both patients. OIH

is part of the spectrum of oesophageal injuries and spontaneous OIH is uncommon. The clinical findings are non-specific and could mimic a wide variety of conditions, but the correct diagnosis could be made with clinical history and characteristic CT findings. CT also delineates the extent of disease, detects possible mucosal or transmural rupture, or other complications such as infection. Although spontaneous OIH is rare, it should be considered a differential diagnosis in patients with acute chest pain and haematemesis. Presentation of spontaneous OIH with haemoptysis is not typical, yet radiologists and clinicians should not overlook this condition as a differential diagnosis.

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