First case of cough-induced combined intercostal artery rupture with partial diaphragm tear

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Abstract

We describe a case of vigorous cough-induced left intercostal artery rupture with partial diaphragmatic tear in a 60-year-old obese male with chronic obstructive pulmonary disease. He presented with left hemothorax, a rapidly spreading chest and abdominal wall hematoma, and progression of anemia. Computed tomography (CT) scan revealed a bleeding focus from the left 8th intercostal artery. CT-guided surface marking of the bleeding point was done over the chest wall. The patient underwent open thoracotomy with drainage of clotted hemothorax and ligation of bleeding intercostal artery. Incidentally, a partial diaphragm tear was detected during the surgery which was repaired. The combined presentation of cough-induced intercostal artery rupture with partial diaphragm tear is not reported earlier. Awareness of these co-existing pathologies can facilitate timely diagnosis and appropriate life-saving management.

Keywords Intercostal artery rupture · Hemothorax · Hematoma · Cough · COPD · Diaphragm

Introduction

Intercostal artery rupture (IAR) is a rare and potentially life-threatening condition [1]. Severe cough is a common accompaniment with acute exacerbations of chronic obstructive pulmonary disease (COPD). We describe a unique presentation in a 60-year-old male with COPD who had vigorous tussive efforts leading to left 8th IAR with a partial diaphragmatic tear. It resulted in a left hemothorax, a rapidly spreading chest and abdominal wall hematoma, and progression of anemia, which was successfully treated surgically with open thoracotomy.

Case report

A 60-year-old diabetic, obese (BMI, 30.2 kg/m²) male with a history of smoking (20 pack years) and COPD (10 years) was admitted to a local hospital with sudden onset severe pain in the left lower chest and abdomen associated with shortness of breath. It started after bouts of severe coughing lasting several

minutes. There was no history of trauma. It was associated with left hemothorax and hematoma over the left chest and abdominal wall. During the course of his admission at the local hospital over 4 days, his hematoma rapidly expanded to involve bilateral abdominal and pelvic walls. His hemoglobin dropped from 14.0 to 9.0 g/dl. He was urgently transferred to our hospital. On arrival, he was tachypneic (respiratory rate, 22/min) with decreased air entry in the left lower zone and bilateral diffuse rhonchi. His vital signs were pulse, 110/min; blood pressure, 100/60 mmHg; temperature, 37.8 °C; saturation, 95% on O₂-4 l/min. Large tender areas of hematoma and ecchymosis were noted over the left lateral chest wall and bilateral abdominal and pelvic walls. His Hb was 8.9 g/dl. Coagulation tests were normal. His spirometry readings were FEV1, 0.89 l (34% predicted); FVC, 1.29 l (39% predicted); and FEV1/FVC, 69% with poor reversibility.

Contrast-enhanced computed tomography (CT) scan of the thorax revealed moderate left-sided hemothorax extending through the 8th intercostal space into the adjoining chest wall soft tissues. A small focal hyperdense focus close to the pleural surface in the region of the 8th intercostal space was present with possibility of active bleeding (Fig. 1). There were no rib fractures. CT scan of the abdomen and pelvis showed diffuse abdominopelvic wall edema. CT-guided surface marking of the hyperdense focus in the 8th intercostal space was performed on the chest wall (Fig. 2).

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Fig. 1 Preoperative computed tomography of the chest showing bleeding of the left 8th intercostal artery with left hemothorax and left chest wall hematoma

He underwent urgent left posterolateral thoracotomy in the 7th intercostal space. Dense adhesions were present between the lung, diaphragm, and the chest wall and adhesiolysis was performed. Large clotted hemothorax was evacuated. On removal of the extrapleural clots, active bleeding from the left 8th intercostal artery was identified. Its location corresponded to the previous CT-guided surface marking on the chest wall. Control of the bleeding was achieved with suture ligation. There was an incidentally detected 3 × 2 cm partial tear of the anterior diaphragmatic muscle fibers which was suture-repaired (Fig. 3). Postoperatively, two units of packed cells were transfused. He required parenteral and later oral steroids along with bronchodilators for adequate control of his COPD. He had significant resolution of the chest and abdominal wall hematoma and was discharged on the 9th postoperative day.

Fig. 2 Preoperative chest, abdominal, and pelvic hematoma with CT-guided surface marking of the site of the bleeding artery

Fig. 3 Intraoperative image showing the partial diaphragm tear

Discussion

This case study discusses an unreported combined occurrence of an IAR with partial diaphragm tear due to vigorous bouts of coughing episodes as an inciting factor in exacerbation of COPD. The patient presented with hemothorax, a massive expanding chest and abdominal wall hematoma, and progressive anemia leading to a potential life-threatening situation.

The most common cause of IAR is trauma [2]. Other underlying etiologies include rupture of intercostal artery aneurysms due to neurofibromatosis type 1 and coarctation of the aorta, anticoagulation use, bleeding disorders, lung infections, and uncontrolled hypertension. Some authors have also reported spontaneous onset IAR without any inciting event such as trauma, non-traumatic arterial wall weakening, or associated illness [1]. The underlying etiology in our case was prolonged forceful coughing due to COPD exacerbation. Severe cough presents in acute episodes of asthma and COPD, generating extremely high intrathoracic pressure differentials which may contribute to serious complications such as pneumothorax, pneumomediastinum, rib fracture, costochondral strain, or cough syncope. IAR due to cough is a very rare complication reported only a few times in literature. Yu et al. [3] have reported the first case of ruptured right 10th intercostal artery with thoracoabdominal hematoma due to violent coughing caused by a viral upper respiratory infection in a 76-year-old hypertensive male. It was successfully treated by transcatheter arterial embolization (TAE). They explained that the cough caused extreme changes in intrapleural pressure to rupture in a vulnerable part of the intercostal artery, especially in a hypertensive elderly male such as their patient. Jang et al. [4] describe a 39-year-old male with severe cough for a few days who had left lateral abdominal wall hematoma induced by rupture of the left 11th intercostal artery. He was

treated immediately by TAE. He was a healthy individual with no comorbid disease apart from obesity (BMI, 33.03 kg/m²). He mentioned that there are changes in the vascularity and the extracellular matrix in the subcutaneous tissue of persons with obesity. The lack of external compression of an abdominal wall hematoma makes bleeding from the intercostal artery progress to severe hematoma. Our patient too was obese which may explain the rapid spread of hematoma arising from the chest wall. IAR can lead to severe hypovolemic shock and early recognition, diagnosis, and rapid treatment remain central for a successful outcome. Durey et al. [5] have described spontaneous bleeding from the 7th intercostal artery immediately after beginning of hemodialysis which was successfully treated with transcatheter arterial embolization. Sundram-Novelend et al. [6] illustrate a case of spontaneous rupture of an intercostal artery aneurysm during air flight which became hemodynamically unstable requiring initial intubation and inotropic support and suggest that prompt intervention is merited whenever these lesions are detected. Our patient showed progression of anemia over 4 days with rapid spread of hematoma over the chest and abdominal and pelvic walls. We also noticed a partial tear of the anterior diaphragm musculature during surgery. The dense adhesions between the chest wall and diaphragm in a patient with chronic COPD could be a potential cause of tear in the diaphragm and the intercostal artery simultaneously during a violent bout of coughing. There was blood oozing from the torn muscle fibers which we suspect may also be the source of hemothorax apart from the ruptured intercostal vessel. The tear may be explained by the sudden vigorous muscular contractions occurring during coughing. Fenner et al. [7] report a case of diaphragmatic rupture after coughing in a 66-year-old male with infection-exacerbated COPD. The initial CT scan was inconspicuous and demonstrated an intact diaphragm. The general condition of the patient worsened and 8 days later, the CT scan was repeated which showed a left diaphragmatic rupture with herniation of abdominal organs into the chest cavity. We suspect our patient could also have possibly developed a delayed diaphragmatic complication if the partial tear in the diaphragm would have been missed since it was not visible in the initial imaging studies and was identified only during thoracotomy.

The optimal strategy of treatment for IAR is still controversial. TAE is a proven effective alternative to thoracotomy [8]. It is the treatment of choice for spontaneous IAR, unless there is a surgical indication [4]. Recent advances in catheter technique may lead to prompt treatment of bleeding and avoid paraplegia by protecting the regurgitation of embolic materials into the radicular and spinal arteries. It is however sometimes difficult to control an active bleeding from small vessels and the catheter manipulation could injure the fragile vessels. One treatment option in our patient could have been to attempt TAE and insert an intercostal drain for hemothorax as done in the case by Mathew et al. [9] who drained 200 ml of fresh

blood. However, we decided to proceed directly with open thoracotomy since we felt that the clotted hemothorax formed over 5 days would not be adequately drained by the intercostal tube and the retained hemothorax could later become infected, possibly leading to the formation of an empyema. We also would be able to control the bleeding vessel through the thoracotomy and hence decided not to perform TAE. In retrospect, we were also able to identify and repair the diaphragm tear which would have gone unnoticed if we had not opened the chest. To our knowledge, this is the first case in the literature describing cough-induced IAR with a diaphragmatic tear. Sometimes a combined approach of TAE and surgery can be taken as documented by Sundram-Novelend et al. [6]. They treated a ruptured intercostal artery aneurysm by therapeutic embolization with Spongostan particles and performed a right thoracotomy to evacuate the clot in the right hemithorax. Conservative management of spontaneous IAR has been reported by Ishida et al. [10] in a 58-year-old male with no associated illness or trauma by blood pressure control and administration of tranexamic acid. They opine that conservative management of IAR may be one of the treatment options after excluding uncontrollable and massive bleeding. Aizawa et al. [11] reported massive and uncontrollable bleeding during surgical repair of the 11th intercostal arterial aneurysm rupture in a patient with neurofibromatosis type 1. It was not easy to identify the precise bleeding point of arterial rupture. They used pressure hemostasis by absorbable oxidized cellulose to control bleeding around the 10th–12th vertebrae. The patient developed paraplegia due to swelling of the cellulose packing in the spinal canal leading to spinal cord compression which was later successfully evacuated with resolution of the paraplegia. We performed a CT-guided surface marking of the bleeding site preoperatively on the chest wall since we anticipated the difficulty in identifying the bleeding point intraoperatively due to the presence of large clots. We noticed the bleeding vessel right below the surface-marked site on the chest wall which led to immediate control of the bleeding vessel.

Conclusion

Cough-induced rupture of the intercostal artery together with diaphragm tear is reported for the first time and is extremely rare. Surgeons and physicians encountering non-traumatic new-onset hemothorax or hematoma formation should consider these conditions as a differential, especially in the context of strong cough due to predisposing conditions like COPD and asthma. Being aware of the co-existence of these pathologies can raise the level of suspicion, facilitate timely diagnosis, and help initiate appropriate life-saving measures for these patients.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Research involving human participants and/or animals None.

Informed consent Obtained.

Consent to publish The participant has consented to the submission of the case report to the journal.

References

1. Dua A, Dua A, Jechow S, Desai SS, Kuy S. Idiopathic spontaneous rupture of an intercostal artery. *WMJ*. 2014;113:116–8.
2. Salmi R, Gaudenzi P, Ricci F, et al. Sudden acute thoracic pain and hypovolemic shock as a presentation of spontaneous rupture of intercostal artery: a case report. *J Am Geriatr Soc*. 2012;60:371–3.
3. Yu WY, Wang CP, Ng KC, Chen WK, Tzeng IH. Successful embolization of a ruptured intercostal artery after violent coughing. *Am J Emerg Med*. 2006;24:247–9.
4. Jang JY, Lim YS, Woo JH, Jang JH. Spontaneous rupture of intercostal artery after severe cough. *Am J Emerg Med*. 2015;33:131.e1–3.
5. Durey A, Kim YS, Kim AJ. Spontaneous intercostal artery bleeding in a hemodialysis patient. *Hemodial Int*. 2017;21:E76–E78.
6. Sundram-Noveland SY, Appleton DS, Screaton NJ, Davies RJ, Chilvers ER. Spontaneous rupture of an intercostal artery aneurysm during air flight. *Thorax*. 2008;63:294.
7. Fenner H, Memeti E, Taha S, et al. Spontaneous diaphragmatic rupture after coughing: report of a case and review of the literature. *Case Rep Surg*. 2019;2019:1270195.
8. Kessel B, Alfici R, Ashkenazi I, et al. Massive hemothorax caused by intercostals artery bleeding; selective embolization may be an alternative to thoracotomy in selected patients. *Thorac Cardiovasc Surg*. 2004;52:234–6.
9. Mathew R, Abdullah S, Renfrew I. Massive abdominal wall haematoma and haemothorax due to spontaneous rupture of an intercostal artery. *Emerg Med J*. 2008;25:608.
10. Ishida A, Hiraoka A, Chikazawa G, Maeda K, Yoshitaka H. Spontaneous intercostal arterial rupture restrained by conservative management. *Ann Vasc Dis*. 2014;7:430–2.
11. Aizawa K, Iwashita C, Saito T, Misawa Y. Spontaneous rupture of an intercostal artery in a patient with neurofibromatosis type 1. *Interact Cardiovasc Thorac Surg*. 2010;10:128–30.

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