Massive hemothorax Caused by Intercostal Artery Pseudoaneurysm: A Case Report.

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Case report

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Abstract

Background: Intercostal artery pseudoaneurysm is rare and at the risk of rupture. The aetiology is always reported to be iatrogenic and traumatic injury. Embolisation is the most common therapeutic method. Here, we report a case of spontaneous intercostal artery pseudoaneurysm and cured by combining covered stent grafting and surgical management.

Case presentation:A 60-year-old man complained of acute right back pain for 5 hours. Computed tomography showed right massive hemothorax and a giant mass with distinct feeding vessel originated from the thoracic aorta within the right hemithorax. Thoracocentesis was performed, and then a covered stent was positioned across the origin of the feeding vessel. The patient was diagnosed with intercostal artery pseudoaneurysm. Finally, we successfully resected the pseudoaneurysm and ligated the proximal part of the artery. Histologic examination have proved the diagnosis. The postoperative course was uneventful, and the patient was discharged on postoperative day 10. There is no recurrence reported during follow-up.

Conclusions: Spontaneous intercostal artery pseudoaneurysm is extremly rare. Delayed hemothorax due to rupture of the pseudoaneurysm may occur years after the formation. Early diagnosis is important and a combined treatment of endovascular intervention and surgical management is feasible, especially for the case of ruptured large tumour-like mass presentation of the pseudoaneurysm.

Background

Intercostal artery pseudoaneurysm is rare and has mostly been described through case reports. Different from the true aneurysm, pseudoaneurysm is a hematoma contiguous with a defect in an arterial wall. It has the risk of rupture and sometimes may cause hemothorax, as in our case, and can be potentially life-threatening. Most of them have been associated with surgical interventions or blunt thoracic trauma. Embolisation is the most common therapeutic method. We report a case of spontaneous intercostal artery pseudoaneurysm and cured by combining covered stent grafting and surgical management.

Case Presentation

A 60-year-old man was referred to our hospital by an ambulance because of acute right back pain for 5 hours. The pain was constant and sharp. Firstly, it was limited to an area just inferior to the tip of his right scapula. But it subsequently radiated to his right chest. There was no associated fever, coughing, shortness of breath, or hemoptysis. He denied any falls or other trauma. He had a history of hypertension for about 4 years without standard treatment and highest reached 180/95mmhg. Chest computed tomography (CT) 3 years prior showed a mass between the right spine and diaphragm measuring 6.4 × 4.3 × 6.3 cm (Fig. 1a). But he did not undergo any kind of medical treatment until this admission. Body temperature was 36.4℃, heart rate was 105 beats/min, blood pressure (BP) was 98/53mmhg, respiratory rate was 20 breaths/min, and oxygen saturation was 96%. Had pallor of conjunctivae. Right-side breath

sounded weaken, left-side was normal. No moist or dry rales. No absence of heart murmurs or muffled heart tones. No midline spine or paraspinal tenderness. Laboratory examination revealed a serum low hemoglobin (74 g/l), high levels of serum D-dimer (6.08 mg/l) and fibrin degradation products (16.50 mg/l). CT showed right massive hemothorax and a 8.1 × 5.9 × 7.5 cm enhanced well-circumscribed mass within the right hemithorax (Fig. 1b). There was a distinct feeding vessel to this mass, which originated from the thoracic aorta (Fig. 1c). Three-dimensional reconstruction of the arterial vasculature suggested a right eleventh intercostal artery pseudoaneurysm (Fig. 2a). We diagnosed that the hemothorax was caused by rupture of the eleventh intercostal artery pseudoaneurysm. Thoracocentesis was performed, and 850 mL of bloody fluid was drained. The symptom of pain was obviously eased and the patient recovered from hemorrhagic shock with blood transfusion. But more than 200 ml/day of blood was drained after the thoracocentesis. On the third hospital day, an operation was planned because of increased hemothorax on chest radiographs and the progression of anemia. Given the large feeding artery emanating from the thoracic aorta, the patient underwent a covered stent grafting to minimize the risk of intraoperative bleeding before the operation. Arteriography showed a tortuous right eleventh intercostal artery with active bleeding in its distal area (Fig. 2b). An endurant II stent graft system ETFC2828C49EE was then positioned across the origin of the right eleventh intercostal artery (Fig. 2c). We subsequently performed a right posterolateral thoracotomy and a giant pseudoaneurysm with huge surface tension was identified. It attached to the right spine through a broad-based pedicle and there was still an active bleeder on its surface (Fig. 3). We found it contains substantial thrombus after an incision was made. The aneurysmectomy and ligation of the proximal part of the eleventh intercostal artery were successful. Intraoperative blood loss was 500 ml. Histologic examination showed that aneurysmal sac was consisted of fibrous tissue with no arterial structure, indicating that it was a pseudoaneurysm. The postoperative course was uneventful, and the patient was discharged on postoperative day 10. He was required to follow-up 1 month later.

Discussion

Communicating with the flowing blood, pseudoaneurysm is a collection of blood outside the vessel lumen [1]. The absence of a 3-layered arterial wall differentiates it from a true aneurysm. Intercostal artery pseudoaneurysms are rare, as far as we know, there are 24 cases reported in the English literature [2–25]. The clinical findings of these cases are summarized in the Table. Ten patients were women and 14 were men. Their age ranged from 9 to 86 years (mean,55). Symptoms were hemothorax in 10 patients, pulsatile mass in 4 patients, hemoptysis in 2 patients, hematoma in 2 patients, acute chest or back pain in 3 patients, and hematemesis in 1 patient. The rest 2 patients without any symptoms were found accidentally by radiological examination. Aetiology of the cases described was iatrogenic in 17 patients, and traumatic in 6 patients. Cause of one patient without any medical history and chest trauma was unclear and ,as well as this case, might be spontaneous.

Hemothorax was the most common presenting symptom in almost all the cases reported previously. So patients with massive hemothorax should be suspected to have an intercostal artery pseudoaneurysm,

especially when they had chest trauma or underwent a surgical procedure via the intercostal space. The rupture of intercostal artery pseudoaneurysm causes

brisk bleeding which may lead to shock or death. But, as reported, delayed hemothorax might occur two to four weeks after trauma or surgical procedures, so early diagnosis was possible and essential. Traditionally, diagnosis of an intercostal artery pseudoaneurysm was usually made by means of arteriography which allows endovascular treatment in a single procedure. But doppler ultrasound and CT are the two most primary diagnostic modality for intercostal artery pseudoaneurysm. In our case, intraoperative findings and pathology on resected tissues were also important in the diagnosis of intercostal artery pseudoaneurysm. Because, as for differential diagnosis, initially, we took pulmonary sequestration and mediastinal tumor into consideration.

The ruptured intercostal artery pseudoaneurysm was a good indication for endovascular intervention. Embolisation was considered to be the preferred treatment of a ruptured pseudoaneurysm. Many successful cases have been reported [8, 11, 14–16, 19, 22–25]. Generally speaking, microcoils were the first choice for embolisation [8, 14, 15, 19, 22, 14, 25], but glue-lipiodol mixture [16], polyvinyl alcohol particles and gelfoam slurry [11, 23] could also serve as alternatives. Callaway et al [5] reported a patient cured with covered stent. Conservative management [6, 20] and ultrasound-guided thrombin injection [9, 13, 17] had also been described. Only few cases involved open excision [2, 4, 7, 10]. Although endovascular intervention was often chosen, sekino et al [8] suggested that a pseudoaneurysm might have multiple blood supplies which sometimes lead to treatment failure. Actually, in our case, the covered stent did not interrupt the blood supply into the pseudoaneurysm completely. Furthermore, atelectasis occured because the thoracic cavity was filled with the hematoma that was difficult to absorb. Surgical removal of the gross hematoma should be performed to prevent infection and release the compressed lungs. So we formulated a two-step therapeutic schedule which was proved to be feasible.

Conclusions

Intercostal artery pseudoaneurysms are rare and have a risk of rupture. Diagnosis before rupture is important. Embolisation is the most common therapeutic method because of its efficacy and low invasiveness, and surgical management is always reserved for embolisation failure. We report a patient with spontaneous intercostal artery pseudoaneurysm. Delayed hemothorax due to rupture of the pseudoaneurysm occurred more than 3 years after the formation. We emphasize the significance of combined treatment of endovascular intervention and surgical management, especially for the case of ruptured large tumour-like mass presentation of the pseudoaneurysm.

Abbreviations

CT
Computed tomography
BP

Blood pressure

Declarations

Ethics approval and consent to participate

Not applicable

Consent for publication

Informed consent for publication was obtained

Availability of data and materials

Not applicable

Competing interests

The authors declare that they have no competing interests

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Authors' contributions

Xiaoliang Li, Gaohua Liu, and Chuanxi Wang participated in the care of the patient

Caiyang Liu, Ran Ran and Ji Li performed the literature review and drafted the manuscript

Caiyang Liu, and Ji Li obtained the image data. All authors read and approved the final manuscript

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Figures

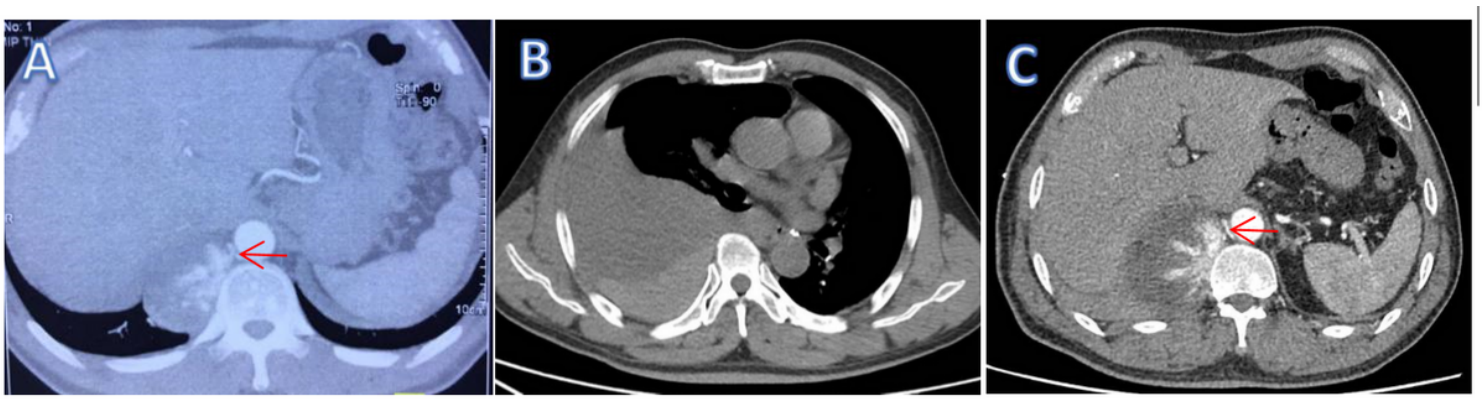

Figure 1

a. Chest CT 3 years prior showed a mass between the right spine and diaphragm. b. Chest CT showed right massive hemothorax. c. Chest CT showed growing mass with distinct feeding vessel.

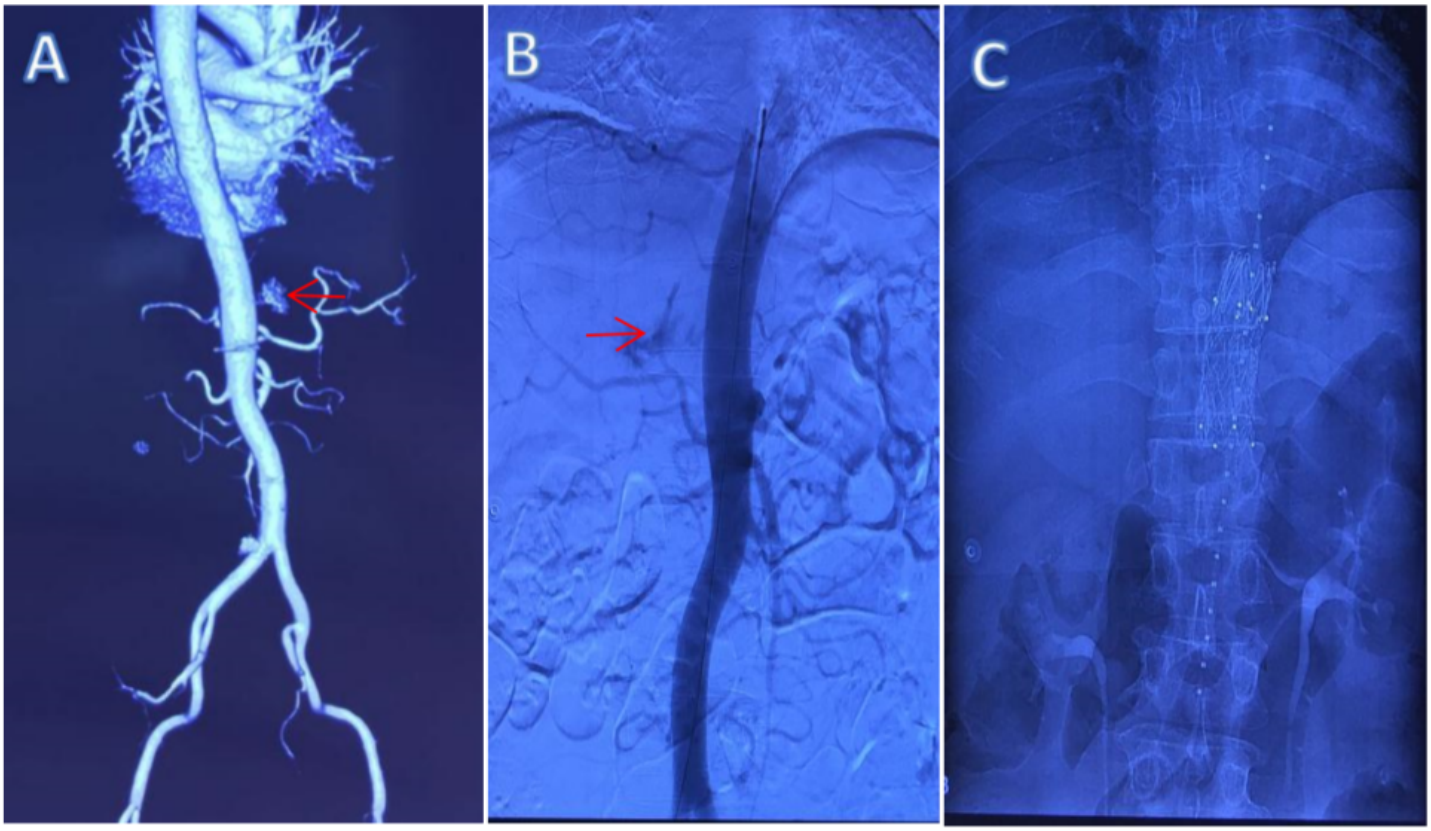

Figure 2

a. Three-dimensional reconstruction of the arterial vasculature. b. Arteriography showed a tortuous right eleventh intercostal artery with active bleeding in its distal area. c. A covered stent was positioned across the origin of the right eleventh intercostal artery.

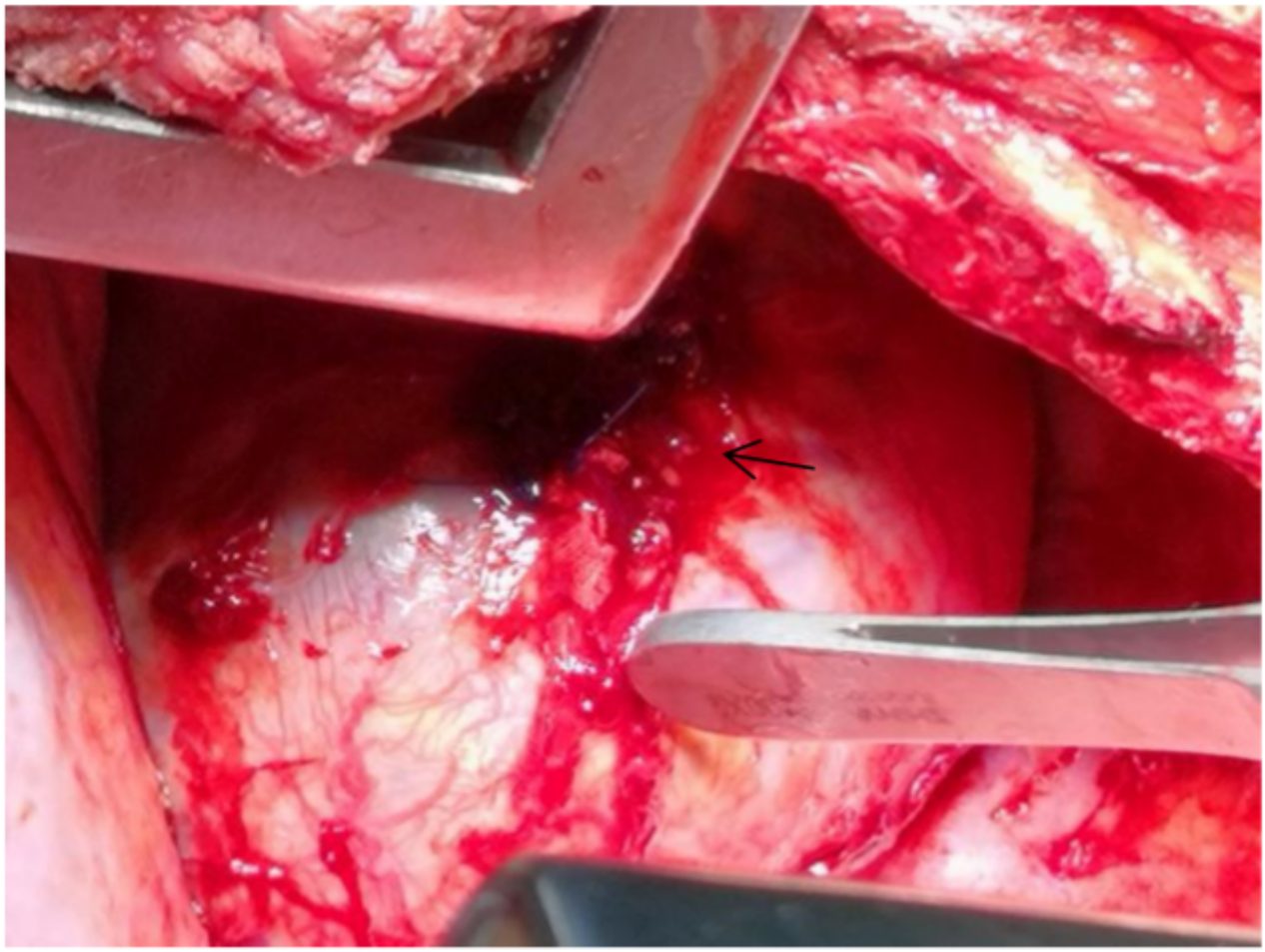

Figure 3

A giant pseudoaneurysm with huge surface tension attached to the right spine through a broad-based pedicle and there was still an active bleeder on its surface.

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- Reportedcasesofintercostalarterypseudoaneurysm.xlsx