Spontaneous rupture of the branches of left subclavian artery

A case report and review of the literatures

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Abstract
Rationale: Spontaneous rupture of the branches of left subclavian artery (LSA) without any obvious risk factors is rare.

Patient concerns: A 51-year-old female patient without history of trauma and hypertension complained about left chest pain.

Diagnoses: A chest Computed tomography (CT) scan revealed a large pleural effusion (PE) in the left thorax cavity and hemothorax was confirmed by thoracocentesis.

Interventions: The patient underwent surgery.

Outcomes: spontaneous rupture of the branches of LSA was confirmed.

Lessons: The patient recovered well and discharged after timely treatments. The unusual possibility should be paid attention in mind in acute chest pain cases.

Abbreviations: ACS = acute coronary syndrome, A-LSA = aberrant left subclavian artery, CECT = contrast-enhanced CT, CT = Computed tomography, HBP = hypertension, KD = Kommerell’s diverticulum, LITA = left internal thoracic artery, LSA = left subclavian artery, PE = pleural effusion, RAA = right aortic arch, RBA = right bronchial artery, RSA = right subclavian artery, VF = ventricular fibrillation, VRD = von Recklinghausen’s disease.

Keywords: chest pain, rupture, spontaneous, subclavian artery, surgery

1. Introduction

Left chest pain is one of the most common chief complaints of the patients in the emergency room. We normally consider cardiogenic etiology, such as acute coronary syndrome (ACS) at initial diagnosis. However, as reported by Conti etc.,[1] among all 6723 patients with chest pain in the chest pain unit, only 1487 (22%) patients were found positive for ACS, and 5236 (78%) patients with non-cardiogenic etiologies. Spontaneous rupture of the branches of subclavian artery is extremely rare, and very few cases were reported in the literature. It is usually associated with primary diseases such as congenital, or acquired vascular anomalies, trauma, tumor, and hypertension (HBP).[2–18] Here we reported a case of spontaneous rupture of the branches of left subclavian artery (LSA) and the patient recovered well by the timely treatments.

2. Case report

A 51-year-old woman was admitted to our emergency service with chief complaint of a persistent pain on the left chest that radiated to the left back and epigastria area. The chest pain had been persisted for 4 hours with chest tightness, sweating, dyspnea, and mild dry cough. There was no palpitation, dizziness, headache, and nausea. The patient claimed a negative history of HBP, diabetes, heart diseases, and respiratory diseases. The left breath sounded low, but no dry and wet rales. This patient was preliminary diagnosed as ACS due to its clinical manifestations. Cardiogenic causes were ruled out by subsequent tests as dynamic 12-lead electrocardiograms, and monitoring of myocardial injury markers. Further observation showed a decrease in systemic blood pressure from 135/80 to 80/65 mmHg accompanied by an increase of heart rate, and the hemoglobin level also decreased rapidly from 11.9 to 6.3 g/dL, indicating the possibility of acute hemorrhage. Forty minutes later, the patient presented signs of compromised hemodynamic parameters. She was pale, and cold, without melena, or haematemesis, but was in abnormal risk. In auscultation, breath sounds could not be heard on the left side of the hemothorax. Computed tomography (CT) (Fig. 1A and B) showed massive pleural effusion (PE) in left thoracic cavity, and unclotted blood was found in thoracocentesis. Subsequently, as hemodynamic parameters continued to deteriorate after fluid resuscitation and blood transfusion, an emergency operation was performed by an exploratory thoracotomy. The patient was prepared on the right lateral decubitus position. Posterolateral thoracotomy was

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performed from the fifth intercostal space. In the left hemithorax, massive hematocoele, clotted blood and hematoma were detected, caused by the multiple ruptures in the branches of LSA. Ruptured vessels were ligated. The patient received 4 units of red blood cells and 860 ml plasma. The hemodynamic parameters were normal during the intra-operative and postoperative period. The blood pressure increased to 123/72 mmHg and the hemoglobin level was 9.1 g/dL. After 6 days of the operation, an angiography (Fig. 2A and B) demonstrated the surgical ligation of the ruptured vessels. Review CT (Fig. 3) scans showed there was no PE about 1 week after the operation. The patient recovered well and discharged on the seventh day after the surgical ligation of the ruptured vessels. No tumor, vascular malformation, aneurysm, and dissection were found in the operation. After 3 months follow-up, there were no long-term complications.

3. Discussion

Spontaneous rupture of the branches of LSA was confirmed in this patient. Cardiogenic diseases were excluded by dynamic 12-lead electrocardiograms and monitoring of myocardial injury markers including myocardial enzymes and cardiac troponin. Then multiple ruptures in the branches of LSA were found during the operation.

However, the biological mechanism of the vessel damage remained unclear. According to clinical practice and reported cases,12–4 HBP is thought to be the main precipitating factor linked to the rupture of vessel. With this specific patient, there
| Series (ref. no.) | Age (yrs) | Sex | Symptoms | CT | Angiography | Basic disease/cause | Treatment | Final diagnosis | Prognosis |
|------------------|-----------|-----|----------|----|-------------|---------------------|-----------|----------------|----------|
| Pentecost 1981[5] | 7         | F   | stridor, dyspnea, numbness of the arm | –  | –           | Neurofibromatosis    | Thoracotomy, grafting | Ruptured aneurysm | Morbid    |
| Takahashi 1989[6] | 38        | F   | hemothorax | –  | –           | VRD                 | Ligation only         | Ruptured aneurysm | Alive    |
| Schievink 1991[7] | 43        | F   | chest pain | hemithorax | left PE | Neurofibromatosis    | Ligation only         | Ruptured aneurysm | Alive    |
| Miyata 1997[8]   | 61        | M   | hemothorax | –  | –           | –                   | –                     | Dissection | Dead     |
| Slisatkorn 2003[9] | 46       | F   | expanding mass and pain, sensory deficit | hemithorax and left PE | Leakage from the LSA | Neurofibromatosis | Ligation only | Spontaneous rupture | Sensory deficit remained unchanged |
| Koh 2005[10]     | 73        | M   | chest and back pain | vascular mass and left PE | – | Neurofibromatosis | Ligation only | Spontaneous rupture | Alive    |
| Yoshida 2005[11] | 48        | M   | shoulder pain and arm paralysis | massive hemothorax | – | Neurofibromatosis | Grafting | Ruptured aneurysm | Alive    |
| Tabelle 2006[12] | 39        | M   | pain from the neck to the left shoulder | aneurysm of the LSA and hemithorax | – | VRD | Ligation, grafting | Ruptured aneurysm | Alive    |
| Matsumoto 2007[13] | 69      | F   | chest pain | fluid in the mediastinum, massive left PE | avulsion of the aorta | pseudo-aneurysm proximal to the A-LSA stemming from a KD | Trauma | Grafting | Ruptured pseudo-aneurysm/RAA | Alive    |
| Bishra 2006[14]  | 40        | M   | chest and back pain | aneurysm of the proximal LSA and hemithorax (CECT) | – | HBP | – | Ruptured aneurysm, aortic coarctation | Died due to VF as a result of renal failure and metabolic acidosis |
| Seow 2007[15]    | 46        | F   | neck pain, expanding mass | mass, LSA aneurysm (CECT) | – | VRD | – | Ruptured aneurysm | Ruptured aneurysm | Alive    |
| Sakamoto 2009[16] | 51       | M   | neck pain, dysphagia | aneurysm of the LSA (CECT) | same findings as CT | Neurofibromatosis | Stent and coil embolization | Ruptured pseudoaneurysm | Alive |
| Taif 2013[17]    | 28        | M   | dyspnea and stridor, progressive oxygen desaturatiion | RAA located at the orifice of an A-LSA, hemithorax and pulmonary (CECT) | same findings as CT | Trauma | Repair | RAA | Died of metabolic disturbance and deranged coagulation function |
| Barbesier 2013[18] | 24       | F   | dyspnea, cyanotic, bleeding from the nose and mouth | – | – | Pregnancy and delivery | – | Dissection | Dead     |
| Shi 2015[19]     | 52        | M   | right chest pain | aneurysm, hemithorax (CECT) | RAA arising from the LSA, an aneurysm at the distal trunk of thoracic RBA | Myocardial infarction, HBP | Percutaneous Embolization | Ruptured aneurysm, RBA | Alive    |
| Present case     | 51        | F   | left chest pain | massive PE in left thoracic cavity | – | Surgical ligature of the ruptured vessels (after operation) | Thoracotomy, ligation | Spontaneous rupture | Alive    |

A-LSA = aberrant left subclavian artery, CECT = contrast-enhanced CT, CT = Computed tomography, HBP = hypertension, KD = Kommerell's diverticulum, LITA = left internal thoracic artery, LSA = left subclavian artery, PE = pleural effusion, RAA = right aortic arch, RBA = right bronchial artery, VF = ventricular fibrillation, VRD = von Recklinghausen’s disease.
was no history of HBP, arteries abnormalities, hereditary diseases as neurofibromatosis or discernible trauma.

An 17 examples of spontaneous rupture of the branches of LSA in patients including ours (Table 1), have been reported in the literature.[6-10] The most common clinical feature of rupture was pain (70.6%, 12/17) with the position of chest, back, and neck, others included sensory deficit (23.5%, 4/17), expanding mass (17.6%, 3/17), dyspnea (17.6%, 3/17), stridor (11.8%, 2/17), hae- morhorax (11.8%, 2/17), and dysphagia (5.9%, 1/17).

Among these 17 patients, we found that spontaneous rupture was happened with congenital or acquired vascular anomalies of the aortic arch, comprised of aberrant LSA itself, such as aneurysms (58.8%, 10/17), right aortic arch (RAA) (17.6%, 3/17), aortic dissection (11.8%, 2/17), and aortic coarctation (5.9%, 1/17).10 of 17 patients with von Recklinghausen’s disease (VRD) (neurofibromatosis) had rupture of LSA, which was considered as mainly hereditary disease that was at risk of rupturing of LSA. 4 patients had a past history of HBP, while one of the patients diagnosed dissection. HBP, no matter acute, or chronic, was very common in LSA dissection, showing that LSA received stronger pulsatile flow than the right subclavian artery (RSA).[12][13][14] Two patients caused by trauma that may have a high mortality. LSA rupture was also reported in postpartum period of pregnancy patient.[4] In pregnancy, and postpartum period, hormonal changes could result in histological changes in the large artery, including fracture of elastic fiber and degradation of acid mucopolysaccharides. However, hemodynamic changes like substantial cardiovascular stress would deteriorate these pathology changes, and increase the risk of dilatation, dissection, or rupture in the large artery.[4]

Chest contrast-enhanced CT (CECT) could provide sufficient and valuable evidences for diagnosis. Among 17 patients, 7 received CECT, and all the results were consistent with the final diagnosis.3 patients received both CECT, and angiography. Angiography should be necessary, especially for the patients who did not receive CECT, because it could be clearer to reveal vascular anomalies, sometimes performed for both diagnostic and treatment purposes.

Spontaneous rupture of the branches of LSA usually needs emergency operation. Among 17 patients, thoracotomy, ligation, repair, and grafting were performed as treatment of the ruptured aneurysm, and ruptured vessels in 12 patients. Two patients received endovascular treatment, which appeared to be a safe and viable alternative to surgery. Usually, early diagnosis, and immediate treatment make good prognosis. Only 2 patients with dissection died of acute massive hemorrhage, and 1 patient with aneurysm died of the upper airway obstruction, without even time for surgery. 2 patients died due to postoperative complications.

4. Conclusion

We reported a case of spontaneous rupture of the branches of LSA without any obvious risk factors as HBP, relevant genetic diseases, artery abnormalities, or discernible wound. It is a rare, interesting, and alerted case, and it reminds us to expand the thoughts of diagnosis in the patients complaining of left chest pain. When a patient complained with chest pain, it is important to be aware of the possibility of aortic or aortic branch rupture if there is no obviously evidence about the respiratory or cardiac diseases.

Author contributions

Formal analysis: J.-K. Jiang, N.-Y. Chen.
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Resources: Y.-Q. Lu.
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Writing – review & editing: Y.-Q. Lu.

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