

## Case Report

# Treatment for unstable pulmonary sequestration injury in patient with severe blunt trauma: A case report

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## ABSTRACT

**Case:** Pulmonary sequestration is a congenital malformation characterized by nonfunctioning tissue not communicating with the tracheobronchial tree. As the blood pressure in the artery feeding the sequestered lung tissue is higher than that in the normal pulmonary artery, the risk of massive hemorrhage in pulmonary sequestration is high. We herein present the first case of a severe blunt trauma patient with unstable pulmonary sequestration injury.

**Outcome and conclusion:** The mechanism of pulmonary sequestration injury is vastly different than that of injury to normal lung. We suggest that proximal feeding artery embolization should be performed before surgical intervention in patients with massive hemorrhage of pulmonary sequestration due to severe chest trauma.

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## Introduction

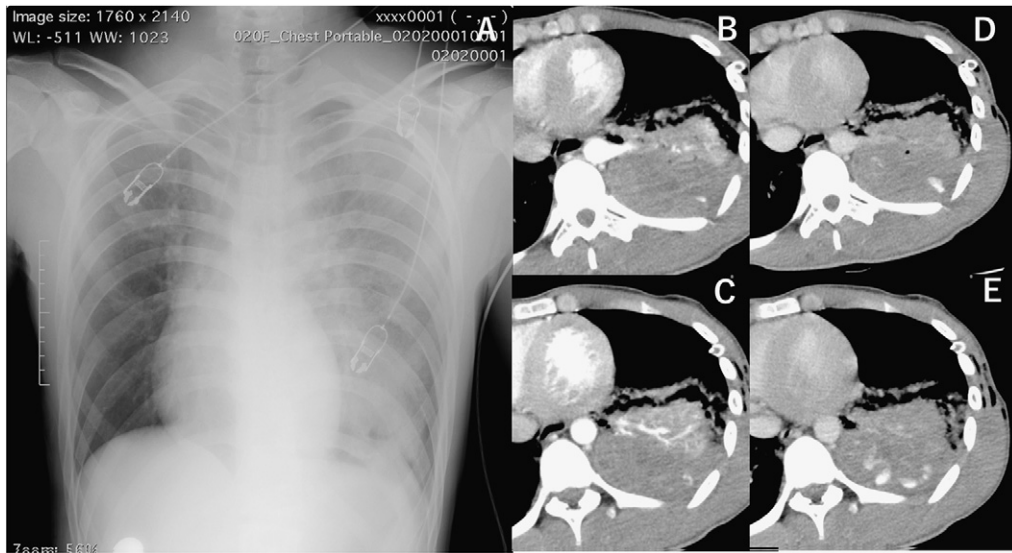
Pulmonary sequestration is a rare congenital malformation characterized by nonfunctioning pulmonary tissue that lacks communication with the tracheobronchial tree and has an anomalous systemic blood supply [1]. As the blood pressure is higher in the artery feeding the sequestered lung tissue than in the normal pulmonary artery, the risk of massive hemorrhage in pulmonary sequestration is high. In fact, massive hemorrhage was previously reported as a life-threatening complication [1,2]. However, unstable massive hemorrhage with pulmonary sequestration injury in a blunt trauma has not yet been reported. We herein present the first case of an unstable patient with massive hemorrhage from undiagnosed pulmonary sequestration injury with a severe blunt trauma.

## Case

A previously healthy 20-year-old man sustained injury due to blunt trauma to the left of his back in a traffic accident. He complained of difficulty in breathing and left-sided back pain on admission. His vital signs were as follows: respiratory rate, 45/min; oxygen saturation, 96% on supplemental oxygen; blood pressure, 100/61 mmHg; pulse rate, 96/min; and body temperature, 37.4 °C. Computed tomography (CT) of the chest and abdomen showed a large, left-sided, basal pulmonary mass with extravasation and multiple rib fractures and burst fracture of the forth lumbar vertebra (Fig. 1). In addition, an aberrant artery 7.6 mm in diameter originating from the descending thoracic artery supplying the mass was observed by CT. Hemorrhagic pleural

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**Fig. 1.** (A) Chest X-ray showing diffuse homogeneous opacities in the entire left lung, indicating pleural effusion in a patient with previously undiagnosed pulmonary sequestration. Note the shift of mediastinum to the right. (B) An aberrant artery arising from the descending thoracic aorta by thoracic computed tomography (CT). (C–E) CT imaging showing active extravasation of contrast medium in the hemorrhagic region of the left lower lobe. A large left hemothorax is also noted.

effusion persisted, and his hemoglobin levels dropped to 7.4 mg/dL. Based on these findings, the patient was diagnosed with pulmonary sequestration with active bleeding.

Although we performed emergency angiography to find the feeding artery that was bleeding to perform superselective embolization, there was no extravasation in selective angiography of the feeding artery (Fig. 2). Therefore, embolization was not performed as the feeding artery had no contribution to massive hemorrhage, and emergency thoracotomy was performed. With the patient in a half-right lateral decubitus position, the laceration of pulmonary sequestration was sutured, and the bleeding was temporarily controlled. However, after the patient was transferred to the intensive care unit, continuous blood drainage from the chest tube was observed over several hours. As the patient's vital signs became unstable, right, one-lung ventilation and massive transfusion were implemented for approximately 8 h, leading to the stabilization of his vital signs. The subsequent clinical course of the patient was uneventful, and he was transferred to the orthopedics ward 18 days after the thoracotomy.



**Fig. 2.** On angiography, an aberrant artery arising from the descending thoracic aorta was observed. Selective angiography of the aberrant artery was performed. No pseudoaneurysm or active bleeding was noted.

## Discussion

In this report, we presented an unstable patient with massive hemorrhage due to undiagnosed pulmonary sequestration injury with severe blunt trauma; the hemorrhage was difficult to control by drainage or/and surgical intervention. Further, we discuss treatment strategies for massive hemorrhage of pulmonary sequestration injury with severe blunt trauma.

The mechanism of injury to pulmonary sequestration is vastly different from that of that to normal lung tissue. First, drainage, suturing, and/or wedge resection may not be sufficient to control bleeding from pulmonary sequestration. Almost 85% of patients with blunt thoracic trauma are managed using conservative approaches, such as drainage, and only 2% of these patients require surgical intervention and/or endovascular arterial embolization; [3] this is due to the low tension in the pulmonary circulation that can terminate the bleeding if hemorrhage is drained from the visceral pleural contact to the parietal pleura [4]. However, the sequestered tissue of pulmonary sequestration is supplied by a systemic blood artery which has a blood pressure is higher than that of the normal pulmonary artery [1]. Therefore, as in our case, bleeding from pulmonary sequestration may be challenging to control by conservative approaches and/or surgical intervention, either of which is usually successful in injury to normal pulmonary tissue.

In addition, emergency surgical resection of pulmonary sequestration is commonly considered as difficult to complete within an hour as the surgical approach relies on identification and control of all aberrant feeding arteries. If all feeding arteries are not properly controlled, life-threatening hemorrhage may occur [6]. Feeding arteries, usually one or two in number, arise from the descending thoracic aorta in 73% of pulmonary sequestration cases. Although less common, subclavian, splenic, or right coronary artery can also be the origin of a feeding artery [3,7]. Due to the variability in the number and origin of feeding arteries among patients, identification of all aberrant feeding arteries by CT or/and angiography is necessary before surgical intervention. Even then, complete control of all feeding arteries may be difficult during emergency surgery that should be completed within an hour.

Moreover, surgical resection is generally indicated for all patients with pulmonary sequestration due to the potential risk of severe recurrent infections, massive hemorrhage, and congestive heart failure caused by left-sided cardiac overload [5], suggesting that it may not be necessary to consider possible complications of complete occlusion of the arterial supply [8]. Recently, endovascular embolization of pulmonary sequestration via embolization of the proximal feeding artery has been reported as a safe alternative to surgery [3,6,8–9]. We did not perform embolization of the feeding artery in this case as extravasation was not observed during emergent selective angiography of the feeding artery; however, similar to other cases with hemorrhage due to pulmonary sequestration, proximal embolization of the feeding artery should be performed regardless of the presence or absence of extravasation by selective angiography of the feeding artery.

There are several limitations to endovascular embolization. The pattern of pulmonary sequestration as well as the number and origin of the feeding artery differ among patients; thus, embolization of the feeding artery is not always easy or feasible. In addition, if the feeding artery creates an artery-to-vein shunt, any obstructing material may reach pulmonary and/or systemic circulation.

In conclusion, the case presented herein illustrates that proximal feeding artery embolization should be performed prior to surgical intervention in patients with massive hemorrhage of pulmonary sequestration due to severe chest trauma.

## Conflict of interest statement

The authors have no conflicts to declare.

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