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ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Dr. Mukul Vij is Senior Consultant Pathologist and Lab Director at Dr Rela Institute and Medical Center in Chennai, India (since 2018). Having received his MBBS degree from King George Medical College in 2004, Dr. Vij undertook postgraduate training at Sanjay Gandhi Postgraduate Institute of Medical Sciences, receiving his Master's degree in Pathology in 2008 and his PDCC certificate in Renal Pathology in 2009. After 2 years as senior resident, he became Assistant Professor in the Department of Pathology at Christian Medical College, Vellore (2011), moving on to Global Health City as Consultant Pathologist and then Head of the Pathology Department (2013). (L-Editor: Filipodia)

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WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Postoperative complications of concomitant fat embolism syndrome, pulmonary embolism and tympanic membrane perforation after tibiofibular fracture: A case report

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Abstract

BACKGROUND

Fat embolism syndrome (FES) is a rare disease characterized by pulmonary distress, neurologic symptoms, and petechial rash and seriously threatens human life and health. It is still neglected clinically because of the lack of verifiable diagnostic criteria and atypical clinical symptoms. No studies on FES with pulmonary embolism (PE) and tympanic membrane perforation have been reported to date. Here, we report a rare case of concomitant FES, PE and tympanic membrane perforation after surgery in a patient with a tibiofibular fracture.

CASE SUMMARY

A 39-year-old man presented with right lower extremity pain due to a car accident while driving a motorbike on the road. X-ray and computed tomography scans revealed a fracture of the right mid-shaft tibia and proximal fibula categorized as a type A2 fracture according to the AO classification. A successful minimally invasive operation was performed 3 d after the injury. Postoperatively, the patient developed sudden symptoms of respiratory distress and hearing loss. Early diagnosis was made, and supportive treatments were used at the early stage of FES. Seven days after surgery, he presented a clear recovery from respiratory symptoms. The outcome of fracture healing was excellent, and his hearing of the left ear was mildly impaired at the last follow-up of 4 mo.

CONCLUSION

Concomitant FES, PE and tympanic membrane perforation are very rare but represent potentially fatal complications of trauma or orthopedic surgery and present with predominantly pulmonary symptoms. Early diagnosis and treatment

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can reduce the mortality of FES, and prevention is better than a cure.

Key Words: Fat embolism syndrome; Tibiofibular fracture; Pulmonary embolism; Tympanic membrane perforation; Postoperative complication; Case report

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Core Tip: Fat embolism syndrome (FES) is a rare complication after internal fixation. This case suggests that any clinical manifestations of patients should be identified after internal fixation to avoid delays in treatment. Even with a lack of verifiable diagnostic criteria for FES, it should be highly suspected for patients with sudden hypoxemia and atypical neurological symptoms. Early diagnosis and supportive treatment are still recommended.

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INTRODUCTION

Fat embolism syndrome (FES) is a rare disease characterized by pulmonary distress, neurologic symptoms, and petechial rash and seriously threatens human life and health. Due to the lack of verifiable diagnostic criteria, the actual incidence of FES varies in different reports^[1-3]. Despite extensive reports of the morbidity and mortality of FES^[4-6], this condition may still be neglected clinically. FES refers to the clinical symptoms that follow a clear history of trauma, particularly long bone fractures (*e.g.*, femur or tibia). Although the exact mechanisms that result in FES are still unknown, it is likely that both mechanical and biochemical components contribute to the pathologic and physical manifestations of FES. Here, we report a rare case of concomitant FES, pulmonary embolism (PE), and tympanic membrane perforation after surgery in a patient with a tibiofibular fracture.

CASE PRESENTATION

Chief complaints

Severe pain and swelling on his right calf.

History of present illness

A 39-year-old man was admitted to the Department of Emergency Orthopedics in our hospital due to right lower extremity trauma from a car accident 2 h prior.

History of past illness

The patient had a free previous medical history.

Personal and family history

The patient had no prior surgeries and did not smoke tobacco or drink alcohol. Family history was negative.

Physical examination

After admission, the patient's temperature was 36.8 °C, heart rate was 89 bpm, respiratory rate was 19 breaths per minute, and blood pressure was 112/89 mmHg. Severe pain and swelling were found on the patient's right calf, while no numbness was found. Peripheral circulation was in good condition, with the dorsalis pedis artery pulse detected. There was limitation of the right ankle joint during flexion and extension. Calcaneal traction was used to relieve pain and reduce swelling after

admission.

Laboratory examinations

There are no specific laboratory tests for FES; however, anemia, hypoxia, decreases in red blood cell count and hematocrit, and fluctuations in the inflammatory indices (*e.g.*, white blood cell (WBC), C-reactive protein (CRP)) were found at the early stage of FES (Table 1).

Imaging examinations

After X-ray and computed tomography (CT) examinations, he was diagnosed with fractures of the right midshaft tibia and proximal fibula (type A2 according to the AO classification) (Figure 1A). Preoperative routine examinations, including chest radiography (Figure 1B) and color Doppler ultrasound of the lower extremity blood vessels, were performed, and the results showed no significant abnormalities.

FINAL DIAGNOSIS

The patient was diagnosed with fracture of the right midshaft tibia and proximal fibula.

TREATMENT

The patient was scheduled to undergo internal fixation of the tibia 3 d after the injury. A successful operation (Figure 1C) was performed *via* a minimally invasive anteromedial approach with an operation time of 90 min and intraoperative bleeding volume of 100 mL.

Unfortunately, approximately 30 h after surgery, the patient developed sudden symptoms of respiratory distress. He became confused and developed tachypnea (respiratory rate of 36 breaths/min) and tachycardia (heart rate: 120 bpm), and his oxygen saturation dropped below 90% on 4 L/min of oxygen *via* nasal cannula. No obvious petechial rash was present. A medical consultation was requested immediately for persistent respiratory distress symptoms, and the patient became weaker and more unresponsive. Decreased breath sounds and a small amount of scattered moist rales could be heard. However, his pupils were equal and round, and his reaction to light was normal. Computed tomography angiography (CTA) of the chest and CT of the head were performed. Moreover, the patient was transferred to the intensive care unit for close monitoring and supportive care. Then, he developed a fever of 38.4 °C 4 h later and required 50 L/min of oxygen to maintain a saturation > 95%. The CT scan of the head did not show any lesions (Figure 1D); however, the chest radiograph (Figure 1E) and CTA (Figure 1F) demonstrated diffuse ground-glass opacities in both lungs, and a limited mural thrombus was found in one distal pulmonary artery (Figure 1G, orange arrow). There are no specific laboratory tests for FES; however, anemia, hypoxia, decreases in red blood cell count and hematocrit, and fluctuations in the inflammatory indices (*e.g.*, WBC, CRP) are found at the early stage of FES (Table 1). Given that the patient's dyspnea was caused by a fat embolism (FE) and PE, high-flow oxygen inhalation (50 L/min), corticosteroids, anticoagulants, and other supportive treatments were used. He experienced hearing loss in his left ear 4 d after surgery, and an otoscopic examination demonstrated that the left tympanic membrane had a small circular perforation (Figure 1H). He was treated conservatively with analgesia and empiric antibiotics.

OUTCOME AND FOLLOW-UP

The patient gradually became more alert, his breathing became smooth, and his hearing in the left ear improved. Seven days after surgery, he presented a clear recovery from respiratory symptoms, showed improvements, and moved all his limbs against gravity without problems. His oxygen requirements decreased from 50 L/min to 4 L/min to maintain a saturation of > 95%. On the 10th d after the operation, the repeat chest CT demonstrated remarkable absorption of the diffuse ground-glass opacities and mural thrombus (Figure 1I and J). The patient returned to the orthopedic ward and was discharged on the 18th d after the operation. The outcome of fracture

Table 1 Laboratory tests

Variable	Reference range	Preoperation	30 h after surgery	48 h after surgery	72 h after surgery
WBC, 10 ⁹ /L	3.50-9.50	13.63	11.25	8.73	14.8
HG, g/L	130-175	146	104	91	93
RBC, 10 ¹² /L	4.30-5.80	4.85	3.27	2.98	2.97
Hematocrit, %	40.0-50.0	44.8	30.5	28.0	28.1
Platelet count, 10 ⁹ /L	125-350	245	198	207	233
CRP, mg/L	0-10.0	8.6	85.4	116.0	44.7
Prothrombin time, s	11.0-15.0	14.0	14.9	14.5	14.5
Prothrombin time/international normalized ratio	0.85-1.50	1.10	1.19	1.15	1.15
Activated partial-thromboplastin time, s	25.0-45.0	37.1	38.1	52.9	39.2
Potassium, mmol/L	3.50-5.10	4.50	3.65	3.76	3.50
Sodium, mmol/L	137-147	141	140	143	141
Chloride, mmol/L	99.0-110.0	104.0	108.6	109.3	108.9
Calcium, mmol/L	2.11-2.52	2.19	1.89	1.95	1.85
Carbon dioxide, mmol/L	22.0-29.0	30.0	27.9	28.9	27.4
Alanine aminotransferase, U/L	9.0-60.0	24.3	70.0	64.0	47.8
Aspartate aminotransferase, U/L	15.0-45.0	21.0	48.0	28.0	29.9
Alkaline phosphatase, U/L	45.0-125.0	49.1	104.0	91.0	58.3
Albumin, g/L	40.0-55.0	42.4	27.8	29.6	31.1
Total protein, g/L	65.0-85.0	68.8	54.5	56.4	51.4
Urea, mmol/L	3.10-8.00	6.19	3.25	5.64	7.63
Creatinine, μmol/L	57-97	62	44	49	60
Uric acid, μmol/L	238-416	311	183	178	163
Arterial blood gas, fraction of inspired oxygen					
Partial pressure of CO ₂ , mmHg	35-48		43	38	37
Partial pressure of O ₂ , mmHg	83-108		58	135	130
Base excess, %	-2.0-3.0		1.3	3.9	2.6

CRP: C-reactive protein; HG: Hemoglobin; RBC: Red blood cell count; WBC: White blood cell count.

healing was excellent, and his hearing of the left ear was mildly impaired at the last follow-up of 4 mo. He was satisfied with the treatments he received.

DISCUSSION

FE is defined as the presence of fat droplets in the systemic circulation after trauma, especially after long bone fracture fixation^[7]. FE is not similar to FES; only a minority of patients develop FES clinically, and most patients present as asymptomatic. Numerous studies have found that early surgical stabilization of the fracture (external or internal fixation) before 24 h dramatically reduced the incidence of FES and pulmonary complications^[9-11]. However, the timing of fracture fixation remains controversial, especially in patients with polytraumatic injuries, in terms of the advantages of early fixation and the risk of serious complications of early definitive fixation (a potential second source of trauma)^[10].

Both mechanical and biochemical components contribute to the physical manifestations and pathology of FES. Mechanically, fat droplets as emboli are forced into the circulation during orthopedic procedures, especially intramedullary fixation. The emboli can affect different organs and cause symptoms, especially in the lungs

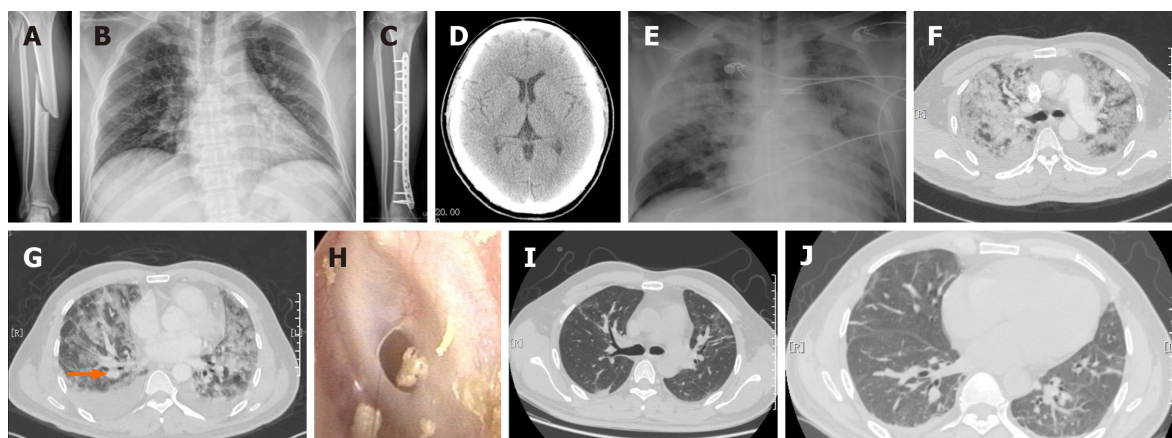


Figure 1 Patient's imaging data. A: Anteroposterior radiograph of the fracture of the right tibia and fibula; B: Anteroposterior radiograph of the chest after admission; C: Anteroposterior radiograph of the right tibia and fibula 3 mo after surgery; D: Computed tomography scan of the brain demonstrated no pathological changes; E: Bedside chest radiography with diffuse interstitial opacities 30 h after surgery; F: Computed tomography angiography of the chest demonstrated diffuse superimposed ground-glass abnormalities in both lungs as well as numerous discrete small nodules, which were consistent with fat embolism syndrome; G: Computed tomography angiography of the chest demonstrated a limited mural thrombus at the right distal pulmonary artery (orange arrow); H: Otoscopic examination demonstrated that the left tympanic membrane had a small circular perforation; I and J: Repeat chest computed tomography demonstrated remarkable absorption of the diffuse ground-glass opacities and mural thrombus 10 d after the operation.

and brain. Biochemically, free fatty acids are released when lipase produced by the lungs acts on embolic fat. Pulmonary endothelial cells and pneumocytes are damaged, leading to arterial hypoxemia^[12].

FES is a potentially devastating cause of morbidity and mortality in polytrauma patients. The clinical features of FES vary. FES usually has an asymptomatic interval for approximately 12 to 72 h after the initial injury, followed by a classical triad of findings: pulmonary distress, neurologic symptoms, and petechial rash^[9]. This condition is often asymptomatic until pulmonary manifestations occur, which is when we first considered FES in this case. A massive pulmonary FE is often found at autopsy. Pulmonary manifestations, including dyspnea, tachypnea, and hypoxemia to acute respiratory distress syndrome (ARDS), are the earliest symptoms and can be seen in 75% of patients^[13]. Hypoxia is the most common finding and is present in 95% of patients^[7]. Neurologic manifestations may include ischemic/hemorrhagic strokes, retinal ischemia, seizures, autonomic dysfunction, and diffuse brain injury^[14], which are present in 80% of patients^[9]. FES is not necessarily accompanied by organic changes in brain tissue; CT scans of the brain may be normal, but brain magnetic resonance imaging is more sensitive in detecting FES^[9,15]. However, isolated cerebral FES without any pulmonary symptoms was reported in a patient with multiple injuries^[16]. This patient initially presented with hypoxia and atypical neurological symptoms 30 h after surgery. The delay in presentation is consistent with the biochemical theory.

A number of respiratory parameters, vital signs, and laboratory results can provide additional information according to Gurd, Schonfeld, and Lindeque's criteria^[9]. Overall, in this patient, we diagnosed FES with PE and conductive deafness based on typical hypoxia (PaO₂: 58 mmHg in the arterial blood), mild neurological symptoms, fever (temperature: 38.4 °C > 38 °C), changes in vital signs (respiratory rate: 36/min > 30/min; heart rate: 120 bpm), and some laboratory tests combined with pulmonary CT findings. Nonetheless, there is no benchmark diagnostic test for FES. Many imaging modalities can facilitate the diagnosis of FES, but none of the findings are specific. Chest radiographic findings may show diffuse bilateral patchy infiltrates, consistent with acute ARDS, although this must be differentiated from pulmonary hemorrhage or pulmonary edema. Computed tomography angiography of the chest can demonstrate diffuse interlobular septal thickening with a superimposed ground-glass abnormality as well as numerous discrete small nodules, which are consistent with FES.

The most important treatment for FES is supportive care for the lung and cardiovascular system to correct hypoxia and maintain hemodynamic stability. Unnecessary transfers between wards and movement should be avoided to reduce the risk of the embolus breaking off and causing cardiopulmonary collapse. If patients exhibit neurologic involvement, frequent neurological examinations and intracranial pressure monitoring should be considered. The use of corticosteroids and heparin has

been suggested as a possible treatment but remains controversial and has not been shown to reduce morbidity or mortality^[17].

CONCLUSION

Concomitant FES, PE, and tympanic membrane perforation are very rare but represent potentially fatal complications of trauma or orthopedic surgery and present with predominantly pulmonary symptoms. This case reminds us to carefully consider and examine similar patients who are encountered in clinical practice. Early diagnosis and treatment can reduce the mortality of FES, and prevention is better than a cure.

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