

Pleural Empyema Concurrent with Arteriovenous Malformation of the Oral and Maxillofacial Regions: A Case Report

Yan Chen¹ | Mengjie Zheng² | Liyan Bo¹ | Congcong Li^{1*}

*Correspondence: Congcong Li

Address: ¹Department of Respiratory and Critical Care Medicine, General Hospital of Northern Theater Command, Shenyang, PR China; ²Department of Maxillofacial Surgery, General Hospital of Northern Theater Command, Shenyang, PR China

e-mail ✉: licong1988@hotmail.com

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ABSTRACT

Introduction: Pleural empyema is a common disease and involves an increasing number of patients. Here, we describe a case of pleural empyema and bacteremia secondary to hemorrhage of arteriovenous malformation (AVM) in the oral and maxillofacial regions.

Case Summary: A 23-year-old female diagnosed with maxillofacial AVM previously presented to our hospital with fever and dyspnea for 8 days. Chest computed tomography (CT) images showed that she had pleural empyema. Bacterial detection showed the growth of *Pseudomonas aeruginosa*, *Actinomyces odontolyticus*, and *Parvimonas micra*. Antibiotic therapy combined with pleural space drainage were not effective until the hemorrhage of the AVM stopped.

Conclusion: AVM rupture and bleeding may lead to persistent or refractory infection, and hemostasis is vital in preventing infection secondary to AVM.

Keywords: Pleural Empyema, Arteriovenous Malformation, Oral and Maxillofacial Region

Introduction

Pleural empyema is the infection and accumulation of pus in the pleural space. According to the most recent estimation, there are over 80,000 cases of pleural infection per year in the USA and UK (Maskell *et al.*, 2006; Farjah *et al.*, 2007). Also, the incidence of pleural empyema increases each year (Stern *et al.*, 2018; Arnold *et al.*, 2021). As research has shown, empyema is a complication that usually follows pneumonia, surgery, trauma, or bronchial obstruction (Arnold *et al.*, 2021; Clark, 2009). For the treatment of pleural empyema, appropriate antibiotic therapy combined with pleural space drainage is effective in most cases.

Arteriovenous malformation (AVM) is a congenital vascular malformation that is characterized by hypertrophied arteries and dilated veins (Ethunandan and Mellor, 2006). These patients can also feel pain in the area surrounding the lesion. It often presents at birth and expands slowly with the growth of

the patient throughout life, which is different from hemangioma (Zhou *et al.*, 2011). AVM can lead to severe complications, such as ulceration, infection, hemorrhage and heart failure, because of the large amount of arteriovenous bypass (Su *et al.*, 2014). However, there was rarely reported about pleural empyema that secondary to AVM.

Herein, we describe a case of a patient who had refractory pleural empyema and recurrent bacteremia because of hemorrhage of the arteriovenous malformation in the oral and maxillofacial region.

Case Report

A 23-year-old female was admitted to the Department of Respiratory and Critical Care Medicine due to fever and dyspnea for 8 days. She was diagnosed with maxillofacial AVM in childhood, and the lesion would intermittently rupture and bleed. The feeding artery of the bleeding site was embolized repeatedly. Eight days before the current admission, the patient had recurrent bleeding from the left gingival lesion where slight pulsations could be palpated. One day later, she developed a fever with a body temperature of 39.0°C, and she also had progressive dyspnea. Blood tests showed elevated leukocyte counts ($23.87 \times 10^9/L$) and procalcitonin (PCT) levels (1.90 ng/mL). The chest computed tomography (CT) results showed shadows of liquid density in the bilateral pleural effusion and part of the lungs (Fig. 1). The pleural effusion of both side was drained, revealing a yellow and turbid pleural fluid with leukocyte count $14 \times 10^9/L$, which indicated pleural empyema. The effusion was determined to not contain malignant cells. Then, she received intravenous antibiotic (cefgixone and metronidazole) treatment. However, her condition deteriorated to tachycardia. Thus, the patient was admitted to our department. Physical examination revealed fever and shortness of breath (body temperature: 38.2°C, heart rate: 132 times/min, respiratory rate: 29 times/min, blood pressure: 153/78 mmHg, and arterial oxygen saturation: 90%). The left gingival lesion was bleeding. CT angiography of the maxillofacial region indicated that there were AVMs on the left side of the face. The left facial artery and superficial temporal artery were the main feeding arteries, and they drained into the left external jugular vein (Fig. 2). Bilateral pleural effusion drainage was performed, and repeated pleural fluid analysis was conducted. The results revealed that the leukocyte count was $12 \times 10^9/L$, lactic dehydrogenase concentration was 1844 U/L, total protein concentration 44.52 g/L, and glucose concentration was 0.78 mmol/L. And the results of the bacterial detection showed the growth of *Pseudomonas aeruginosa* (Day 7, microbial whole-genome sequencing of pleural fluid), *Actinomyces odontolyticus* (Day 7, blood culture) and *Parvimonas micra* (*P. micra*) (Day 21, microbial whole-genome sequencing of pleural fluid). The patient was administered empirical initial antibiotic treatment, and the antibiotics were adjusted according to the pathogenic detection results (Fig. 3). Repeated cardiac and abdominal ultrasound

examination showed no abnormalities. However, the patient remained febrile (38.7 °C), leukocyte count was $21 \times 10^9/L$ and PCT concentration was 1.85 ng/mL. On the 32 day, the bleeding stopped after using hemostatic drugs including aminocycloacid and restore coagulation factor by transfusing of fresh frozen plasma. We continued to give her intravenous antibiotic therapy combined with pleural space drainage, and the body temperature returned to normal. The elevated leukocyte count and PCT also dropped to normal on Day 37 (leukocyte count was $8.0 \times 10^9/L$ and PCT was 0.05 ng/mL). These indicated that the bleeding of the lesion and changes in the patient's condition, such as fever, leukocyte count and PCT, were synchronous.

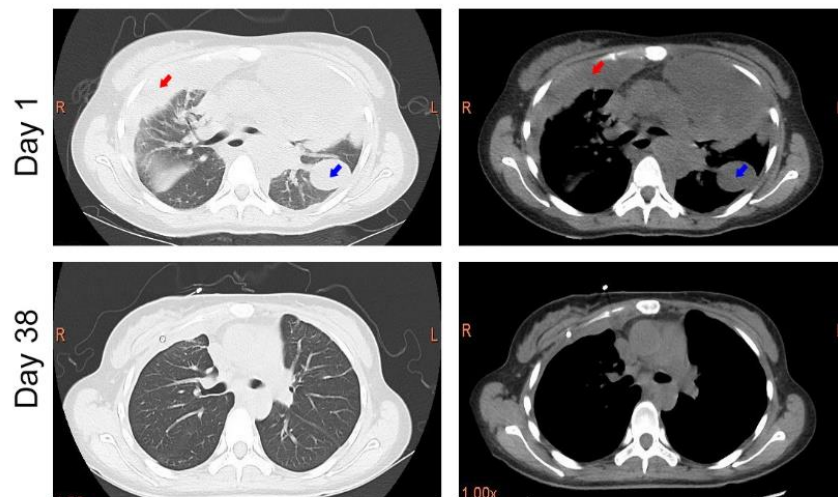


Figure 1: Chest computed tomography. The chest computed tomography results showing shadows of liquid density in both lungs (blue arrow) and bilateral pleural effusion (red arrow).

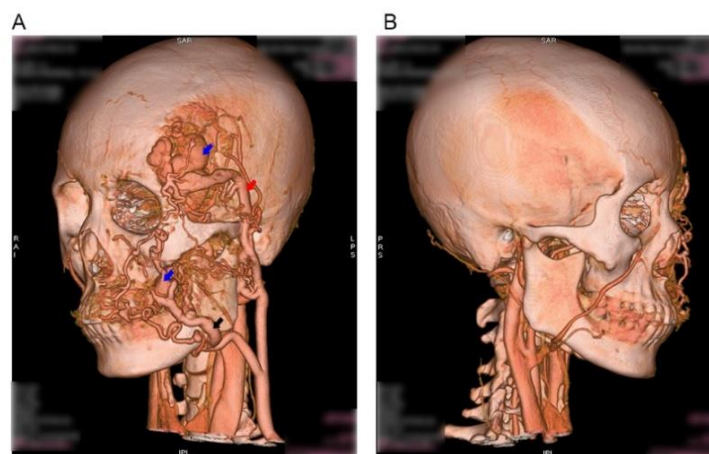


Figure 2: Computed tomography angiography of the maxillofacial region. Computed tomography angiography of the maxillofacial region indicating the presence of hemangiomas on the left side of the face (blue arrow). The left facial artery (black arrow) and superficial temporal artery (red arrow) were the main feeding arteries, and they drained into the left external jugular vein.

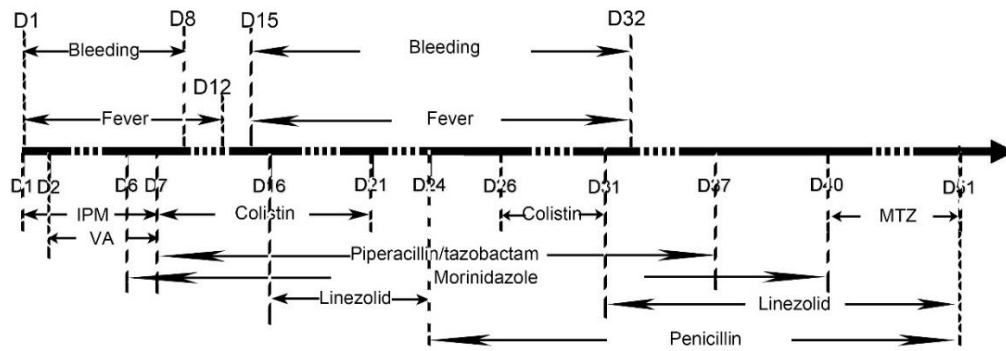


Figure 3: Time sequence of the patient's condition and therapeutic schedule. MTZ: Metronidazole; VA: Vancomycin; IPM: Imipenem Cilastatin.

Discussion

The annual incidence of pleural empyema is 10-12 per 100,000 inhabitants (Farjah *et al.*, 2007). Diabetes mellitus, alcohol abuse, poor dental hygiene, gastroesophageal reflux, chronic lung disease, and intravenous drug abuse might increase its incidence (Farjah *et al.*, 2007). According to research conducted in the United States of America and United Kingdom, the mortality rate is approximately 10-15% (Maskell *et al.*, 2006; Farjah *et al.*, 2007; Arnold *et al.*, 2021). The standard treatment protocol is appropriate antibiotic therapy combined with pleural space drainage. In a survey including 261 pleural empyema patients, 64% had gram-positive cocci, with the most common being *Streptococcus pneumoniae* (Falguera *et al.*, 2011). Furthermore, 10% of these patients had anaerobes, 6% had gram-negative organisms, and only 4% had atypical organisms. As the pathogenic bacteria vary, empirical initial antibiotic treatment followed by antimicrobial treatment according to the underlying etiology is essential (Stern *et al.*, 2018; Godfrey *et al.*, 2019).

AVM is a rare congenital vascular deformity. Whereas hemangiomas, which are often confused with AVM, occur at birth or soon after birth and often have a rapid proliferative phase and an involution phase (Perez *et al.*, 2010). Patients with AVM often present with a high skin temperature, and sometimes vascular noise can be heard or arterial pulsation can be felt. AVM is difficult to treat, but many treatment methods have been evaluated. Ligation or embolization of the feeding artery often aggravates the disease because it can lead to ischemia and necrosis of the parenchyma (Su *et al.*, 2014). With the establishment of collateral circulation, the lesion will eventually recur and sometimes become even more severe (Su *et al.*, 2014). Excising the lesion by surgery thoroughly is an option. For lesions not involving the intracalvarium, dehydrated alcohol injection can be effective; however, this method may also sometimes lead to necrosis of the parenchyma.

The current patient successively became infected by three types of bacteria: *Pseudomonas Aeruginosa*, *Actinomyces odontolyticus* and *Parvimonas micra*. *Actinomyces odontolyticus* was first isolated from persons with advanced dental caries. According to a previous study (Brailsford *et al.*, 1999), it could be isolated from the dentine of 90% of subjects. These bacteria are often found in the cervicofacial regions, chest, abdomen and pelvis. *Parvimonas micra* is a gram-positive anaerobic coccus that is frequently isolated from dental plaque in patients with chronic periodontitis. It is a common constituent of mixed anaerobic infections. This bacterium was also implicated in infections associated with recent dental procedures. As two of the three types of infected bacteria are common bacteria in the oral cavity, we speculated that the source of pleural empyema and bacteremia was AVM lesions in the oral cavity. Bleeding lesions of the left gingiva easily spread the bacteria. Moreover, the bleeding of the lesion and changes in the patient's condition, such as fever, leukocyte count and PCT, were synchronous. Therefore, we deemed that the patient had refractory pleural empyema and recurrent bacteremia because of hemorrhage of the AVM in the oral and maxillofacial regions and revealed the risk of infection with an AVM in the oral cavity.

Conclusions

We describe a case of pleural empyema and bacteremia secondary to hemorrhage of AVM in the oral and maxillofacial region and revealed the risk of infection with an AVM in the oral cavity. The rupture and bleeding of AVMs may lead to persistent or refractory infection; thus, hemostasis is also vital in preventing infection secondary to AVMs.

Abbreviations

PCT: Procalcitonin

CT: Computed Tomography

MTZ: Metronidazole

VA: Vancomycin

IPM: Imipenem Cilastatin

AVM: Arteriovenous Malformation

Authors' Contributions

Y.C. and L.Y.B. contributed to the data collection and manuscript composition. M.J.Z. and C.C.L. contributed to the case presentation and manuscript composition. C.C.L. was responsible for the integrity of this work. All authors contributed to drafting the manuscript and have read and approved the final manuscript.

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