Lemierre's Syndrome Presenting as Acute Respiratory Distress Syndrome after Palpitis Odontogenic Infection: A Case Report

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ABSTRACT

Introduction: Lemierre's syndrome, is one of forgotten diseases primarily caused by anaerobes Fusobacterium species, with possible other unusual organisms. It occurs after the extension of oropharyngeal infection into pharyngeal spaces of the neck with subsequent septic embolism to different parts of the body.

Case: A case described is of a 36-year-old male who presented with neck pain and swelling and five weeks later developed fever, hemoptysis and acute respiratory distress syndrome. A chest radiograph showed diffuse bilateral infiltrate with consolidation and CT-scan chest with extension to the neck revealed dilated IJV with soft tissue swelling. Sputum culture was positive for Pseudomonas aeruginosa, rarely implicated to cause Lemierre's syndrome. The patient was treated with anticoagulant and intravenous antibiotic therapy with a good response although developed dysphagia as a complication.

Conclusion: Lemierre's syndrome can present atypically with ARDS. High index of suspicion is needed with assistance from microbiological and radiological investigations. Management can be challenging in low resource setting causing major implications in antimicrobial stewardship and proper antimicrobial use, management costs and patient outcome.

Keywords: Lemierre's syndrome; Septic embolism; Antimicrobial stewardship

Abbreviations; CT- Computed Tomography, IJV- Internal Jugular Vein, ARDS- acute respiratory distress syndrome, HR- heart rate, BPM- beats per minute, BP- blood pressure, RR- Respiratory rate, IV- Intravenous, WBC- White blood count, CXR- Chest X-ray, NGT- Nasal gastric tube, OGD- Oesophagogastroduodenoscopy,

INTRODUCTION

Lemierre's syndrome, is one of the forgotten diseases by many physicians.[1] It is a rare complication named after the French physician Andre Lemierre, which arises from bacterial pharyngitis/tonsillitis and rarely from the oral cavity.[2] It is caused by the extension of infection into the lateral pharyngeal spaces of the neck with subsequent septic thrombophlebitis of the internal jugular vein(s), leading to pneumonia or pleural effusion.[3] The most common implicated organism causing Lemierre's syndrome is Fusobacterium necrophorum, followed by other Fusobacterium species such as F. nucleatum, F. mortiferum and F. varium. [4] This is a case report of a 36-year-old male presenting with odontogenic infection leading to Lemierre's syndrome which presented as ARDS, pneumonia and later leading to dysphagia.

CASE PRESENTATION

A 36-year-old man came to the dental clinic complaining of painful swelling on the right side of the lower jaw and neck for one week following painful tooth on the same side for 1 month with difficulty in opening

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mouth and painful swallowing both solids and watery food. Physical examination revealed body temperature of 36.9^0C, regular heart rate (HR) of 122 BPM, respiratory rate of 20 breath cycles per minute, blood pressure (BP) measured was 100/60mmHg and oxygen saturation (SpO2) of 96% in room air. He was reviewed by dental surgeon and was found to have facial asymmetry with diffuse, firm and tender swelling on the right side of the lower jaw that included submandibular region, extending to the angle of the mandible and temporomandibular joint. An intraoral examination revealed mild trismus and pus oozing through the mouth. The patient was diagnosed as a case of odontogenic infection (Pulpitis) complicating to dental micro abscesses and Ludwig's angina.

He was admitted in the surgical department and planned for incision and drainage and was kept on empirical intravenous antibiotics (IV Metronidazole and IV ceftriaxone, analgesic (IV diclofenac) for 17 days and later switched to oral medication. Multiple incision and drainage were done, with daily dressing. Investigations on admission showed Full blood count WBC= $14x10^9$/L with neutrophilia ($12.7x10^9$/L), normal platelets and hemoglobin level, serum creatinine= 85mmol/l, and normal chest X-ray (as shown in Figure 1).

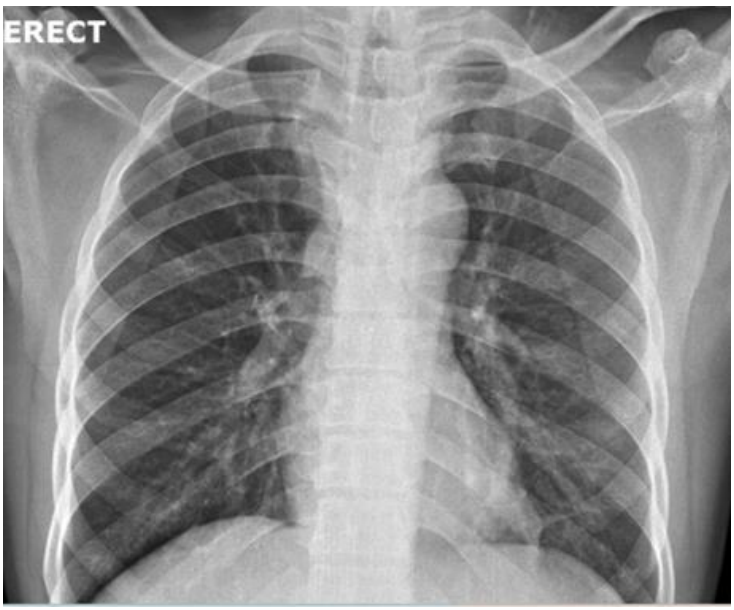

Figure 1: Posterolateral Chest X-ray during admission which shows normal findings

Five weeks later (since his admission in the surgical ward), he still had neck pain and swelling and developed acute coughing blood and progressively and rapidly worsening difficulty in breathing, high grade fever, chills and excessive sweating in the evening. On physical examination: he was febrile with temperature of 38.8ºC, BP= 132/74mmHg, PR= 96bpm, RR= 28 breaths cycles per minute and Oxygen saturation (SpO2) = 88% on room air. Respiratory system examination revealed bilateral coarse crepitations all over the chest. The diagnosis of acute respiratory distress syndrome (ARDS) secondary to pneumonia with possible septic embolization from the dental abscesses/periodontal abscesses was suspected. The patient was transferred to the medical department.

Repeat blood analysis revealed: Leucocytosis of 51.25 $x10^9$/L with neutrophilia ($49.97x10^9$/l), Hemoglobin= 9.7g/dL, Platelets= $640x10^9$/L, Malaria Rapid Diagnostic Test- Negative, sputum for Gene Xpert was negative for Mycobacterium Tuberculosis (2 samples), Sputum Gram stain showed Gram negative rods and sputum culture revealed Pseudomonas aeruginosa sensitive to Amikacin, Ceftazidime and Piperacillin/Tazobactam. Repeat CXR (as shown in Figure 2) showed features suggestive of pneumonia and right-side effusion. CT scan of Chest was done and showed in-homogeneous opacity with multiple air bronchogram seen within right middle and upper lung segments associated with loss of lung volume suggestive of Pneumonia with right pleural cavity effusion and ruled out pulmonary embolism. Exposed part of the neck showed a dilated IJV, soft tissue swelling with multiple air pockets suggestive of thrombophlebitis and soft tissue inflammation (Ludwig's angina) respectively.

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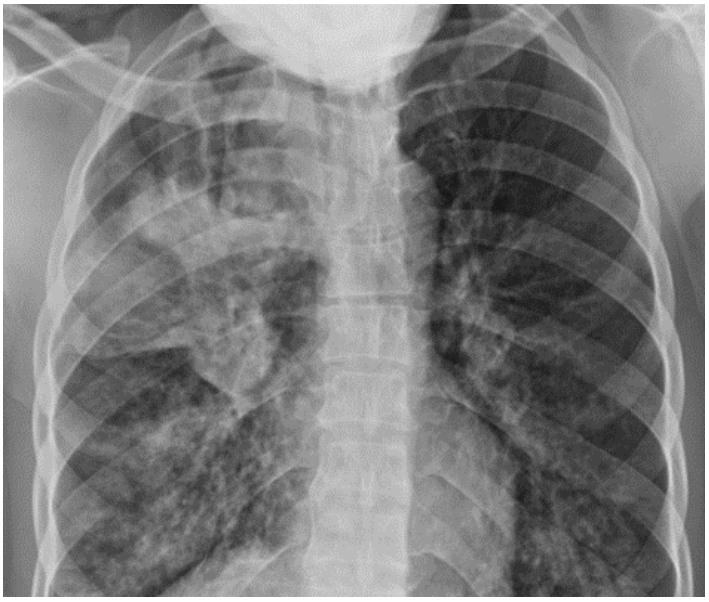

Figure 2: Posterolateral chest x-ray: 5 weeks later showing homogenous opacity with air bronchograms seen in upper and middle lung zones, patchy opacities in lower zones, bilateral reticular nodular infiltrates with homogenous consolidation of upper right lobe

He was kept on oxygen therapy 15l/min through non-rebreather mask, and was given IV dexamethasone injection. Antibiotic treatment was changed to IV Piperacillin/Tazobactam 4.5g three times a day for 15 days and started anticoagulation with Tabs Rivaroxaban 15mg od for 1 week. Oxygen therapy was weaned off after 3 days as he had shown significant improvement. During the course of treatment, difficult and painful swallowing worsened and was unable to swallow both liquids and solids, which necessitated insertion of a nasogastric tube with difficulty. He continued to feed through NG tube for 1 month due to difficulty swallowing even after trying to remove it. Oesophagogastroduodenoscopy (OGD) and repeat/post treatment x-ray could not be done due to socioeconomic issues. He stayed in the medical ward (16 days) with total hospital stay for 52 days and was discharged in good condition with oral antibiotics.

DISCUSSION

Lemierre's syndrome is most commonly caused by the anaerobic normal flora of the oral cavity and neck.[1, 3] There are other bacteria which have been rarely found and reported to cause the syndrome which include Bacteriodes, Eikenella, peptostreptococcus, Streptococcus, Proteus and Staphylococcus aureus.[5] Contrary to our patient, the septic embolic pneumonia he developed was caused by Pseudomonas aeruginosa which is less implicated as the cause of Lemierre's syndrome. Anaerobic blood culture could not be done due to unavailability of anaerobic culture bottle and media in our hospital; hence it is difficult to conclude total absence of the most common anaerobe (i.e. Fusobacterium necrophorum) as the cause, or a possibility of presence of multiple causative organisms.

This condition can be difficult to diagnose due to atypical presentation which requires high index of clinical suspicion, usually presenting with nonspecific symptoms which may include fever, neck pain, neck swelling and odynophagia.[6] The common presentation of septic embolism is pneumonia and pleural effusion.[3] Although there are other less common presentations such as soft tissue abscesses, brain abscess, spleen and liver abscesses, osteomyelitis, endocarditis and pyomyositis.[3, 7] Some patients present with ARDS, similar to how our patient presented.[8]

Contrast-enhanced computed tomography of the neck is the best diagnostic method.[3] Our patient performed a CT scan of the chest with extension to the neck, which revealed soft tissue swelling around the neck, dilated IJV and multiple consolidation in the lung with mild pleural effusion. Persistent neck swelling, pain and dilated IJV on CT scan led to suspension of possible presence of thrombophlebitis, meanwhile the

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development and fast progression of respiratory symptoms and the radiological findings confirmed our suspension of Lemierre's syndrome. Evidence from previous literature have reported the diagnosis of the syndrome supported by clinical and other radiological features even in absence of radiological evidence of thrombus/thrombophlebitis in the IJV. [9]

This patient was admitted and given IV antibiotics for two weeks without improvement. During this time, no culture samples were obtained until the patient experienced fever and respiratory distress. This has significant implication in antimicrobial stewardship and medical costs. Since this patient had clear infection in oral cavity with multiple abscesses, pus and blodd culture samples should have been taken before starting the IV antibiotics, and the results would have helped with appropriate choice for treatment: and would have likely avoided this complication if he were to be treated with appropriate antibiotics.

Lemierre's syndrome causes different complications to patients such as stroke, vocal cord palsy, bronchopleural fistula to mention a few.[7] Our patient developed dysphagia, which is a rarely reported complication of Lemierre's syndrome. This could have been caused by extensive soft tissue inflammation around the neck structures which might had involved the oesophagus with presumed fibrosis.

Management of Lemierre's syndrome can be challenging especially in low resource setting, such as in many countries in Sub-Saharan Africa. It can be very costly and requires extensive and advanced investigations such as CT scan, microbiological workup and expensive medication including anticoagulation and prolonged antibiotics use. At times, it requires multidisciplinary team approach with prolonged hospital stay. Availability of such conducive infrastructure is a major challenge in improving overall care especially in many health facilities in low resource setting.[10]

CONCLUSION

Lemierre's syndrome carries high mortality if not treated on time and requires high index of clinical suspicion. Meanwhile, proper antimicrobial stewardship measures in managing oral pharyngeal infections are required not only to prevent this condition, but also to reduce development of antimicrobial resistance. This will also reduce socio-economic burden in treating our patients by reducing long hospital stays, improving quality of care and reducing mortality.

Consent:

A written inform consent was sought from the patient before publishing this case.

Conflict of Interest:

There is no conflict of declaration in publishing this case.

Funding:

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