IDCases 10 (2017) 15–17

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcases

Case report

Lemierre's syndrome: Case report and brief literature review

Andrew Zhao[a], Mohammed Samannodi[b], Muhammad Tahir[b], Sarah Bensman[b], Michael Hocko[b]

[a] Mayo Clinic Health System – Franciscan Healthcare, 815 10th Street, South La Crosse, WI, 54601, United States
[b] Department of medicine, Catholic Health System, University at Buffalo

ABSTRACT

Lemierre's syndrome has been shown to be increasing in incidence in the past 20 years with one popular suggesting that said rise occurred from less aggressive antibacterial coverage. We report a case of Lemierre's syndrome and also reviewed the 15 most recent case reports. A previously healthy 25 year old male who initially developed sore throat and flu-like symptoms, was prescribed antibacterials as an outpatient but was hospitalized for worsening symptoms. He was later diagnosed with Lemierre's syndrome and improved clinically with IV antimicrobials alone. From our concise literature review, we determined that a decrease in antibiotic prescriptions may not fully explain why the incidence of Lemierre's has been increasing. Thus, future research should be focused in evaluating possible worsening susceptibilities to antibiotics and improvements on detection. We also advise physicians to be aware of the signs and symptoms of this rare but potentially fatal condition as well as the available detection methods and treatment

Introduction

In 1936, Andre Lemierre reported 20 young, previously healthy young adult patients who were initially diagnosed with pharyngotonsillitis or peritonsillar abscess and developed neck swelling and tenderness secondary to septic thrombophlebitis of the internal jugular vein, metastatic abscesses, and anaerobic septicemia, leading to the death of 18 patients [1]. This constellation of findings, now named Lemierre's syndrome, had decreased in prevalence since the advent of antibiotics but is now being reported more frequently, especially in the past 20 years [2]. One of the proposed explanations for this phenomenon is that antibiotic prescription practices have become more conservative, resulting in more patients who are not being treated for their bacterial infections and are now more susceptible to developing Lemierre's syndrome as a complication [2]. We report a case of a previously healthy young adult male who was initially given antibiotics but later returned with worsening symptoms requiring hospitalization. We also performed a brief literature review of the most recent 15 published cases of Lemierre's syndrome with the patients' initial symptoms, if they saw a healthcare practitioner initially, and if they were prescribed antibiotics.

We present a 25 year old male who presented to ED with a 4 day history of severe sore throat, mild diffuse abdominal pain and flu-like symptoms of body aches, weakness, and fever. He had originally visited an urgent care center 2 days prior, where Monospot and Rapid Strep tests were negative. He was discharged at that time with azithromycin and prednisone. His symptoms progressively worsened and he also reported new onset nausea, vomiting, diarrhea, dizziness, sweating, chills and 2 episodes of syncope, which prompted this visit to the ED. He denied any recent dental procedures, sick contacts, recent travel, or IV drug use.

His physical exam at that time revealed slightly low but still normotensive blood pressure of 104/60 mmHg, tachycardic heart rate at 133 bpm, respiratory rate 17 bpm, temperature 101.7 F, and oxygen saturation at 93% room air. Significant findings included tonsils with 3+ enlargement, erythematous posterior pharynx with no exudates, dry mucous membranes, and positive lymphadenopathy of anterior and posterior cervical lymph nodes. His chest was clear to auscultation and percussion bilaterally with no tenderness of the chest wall. Exam of his skin did not show any Janeway lesions, Osler nodes or splinter hemorrhages

A complete blood count showed significant leukocytosis with WBC 35,400, hemoglobin 13.5 g/dL, hematocrit 39.9%, and platelets 557,000. He was found to have mild hyponatremia with a sodium of 131 mEq/L but with otherwise normal electrolytes (potassium 5 mEq/L, chloride 95 mEq/L, CO2 25 mEq/L), normal kidney function (BUN 12 mg/dL, creatinine 0.85 mEq/L). Liver function was found to be abnormal with ALT 103 IU/L, AST 48 IU/L, alkaline phosphatase 122 IU/L, total bilirubin 2.0 mg/dL. Initial imaging consisted of a chest x-ray which showed no acute cardiopulmonary process. (Fig. 1) A CT-angiogram of the chest with and without contrast showed multiple cavitary lung masses, suggestive of septic emboli, including a dominant cavitary lung mass at the right base measuring up to 4.4 cm. There was also a trace right pleural effusion. (Fig. 2) Tuberculosis testing was

E-mail address: zhao.andrew@mayo.edu (A. Zhao).

http://dx.doi.org/10.1016/j.idcr.2017.07.009
Received 4 June 2017; Received in revised form 23 July 2017; Accepted 24 July 2017

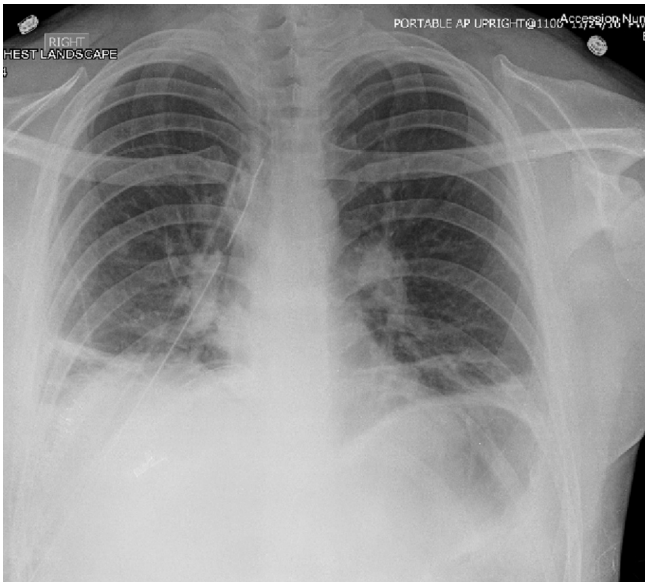

Fig. 1. Chest x-ray.

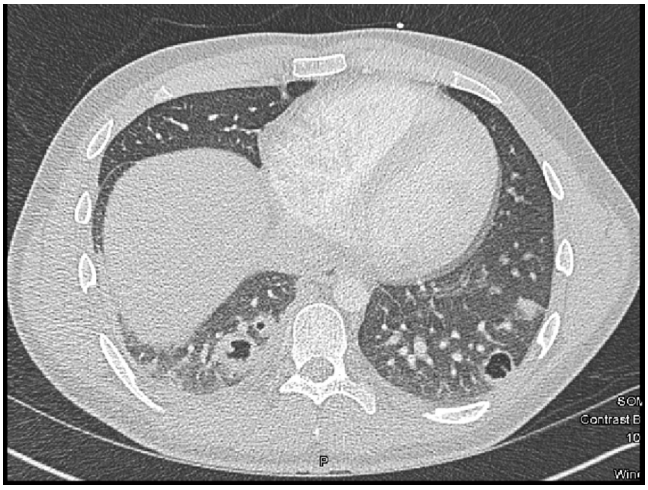

Fig. 2. CT angiogram with contrast showing new multiple cavitary lung masses, suggestive of septic emboli, and trace right pleural effusion.

performed as well and found to be negative.

Patient was subsequently admitted under the Infectious Disease service for diagnosis of septic emboli. He was placed on IV piperacillin/tazobactam and IV vancomycin. Echocardiogram showed normal left ventricular ejection fraction at approximately 65 +/− 5%, mildly enlarged right ventricle. No clots, masses or vegetations were visualized. The patient underwent a right video-assisted thoracoscopic surgery with drainage of pleural effusion (400 mL), partial decortication, and right lower lobe wedge excision of right lower lobe lung abscess by cardiothoracic surgery. Lung tissue and pleural fluid cultures were obtained. There was concern for Lemierre's disease so a Doppler of bilateral internal jugular veins was obtained, which was negative for thrombus. Carotid Dopplers were also performed and showed mild stenosis (0–50%) in internal carotid arteries bilaterally and normal vertebral flow bilaterally. On the 5th day of incubation, the culture of the right lower lung tissue grew fusobacterium necrophorum. Blood and pleural fluid cultures returned negative. Lung tissue pathology for the wedge resection showed acute bronchopneumonia with abscess formation, rare blood vessels with organizing thrombus and focal infarction with no evidence for malignancy.

The patient's clinical course continued to improve; WBC count trending down to normal range and his fevers resolved. A PICC line was placed, antibiotics were de-escalated to ceftriaxone and metronidazole and the patient was discharged home. At this time, patient has continued to do well with no recurring symptoms.

Discussion

Our case is unique in that the patient did not meet the usual diagnostic criteria for Lemierre's syndrome, which includes positive blood cultures and radiological evidence of internal jugular venous thrombophlebitis. However, our patient's clinical course strongly suggested Lemierre's syndrome in that he initially presented with an oropharyngeal infection which progressed to sepsis and developed septic emboli growing fusobacterium necrophorum, a microbe that is not part of normal oropharyngeal flora and closely associated with past cases of Lemierre's. Riodan et al. (2004) performed a literature review involving 5 case series and analyzed the major features found. IJV thrombosis, one of said features, was found to be present in only 26–45% of all cases while anaerobes grew from blood cultures in 97–100% of those cases. [3] However, Eilbert et al. (2013) presented a patient who did not have positive blood cultures. In addition, they also referenced other cases with negative cultures and discussed the possible causes of this diagnostic outcome, including length of time needed for cultures to grow and suppression of growth because of prior antibiotic use, which would include our patient. [4] As there is no set standard diagnostic criterion for Lemierre's syndrome, we strongly recommend that providers rely on their clinical judgment especially considering the rarity of this condition and possible variants.

Physicians have noticed a resurgence of Lemierre's syndrome in the past recent years. One of the proposed hypotheses for this phenomenon is the trend against prescribing antibiotics for sore throats, leading to more cases developing complications that would have been prevented otherwise. In order to evaluate that claim, we reviewed 15 of the most recent published case reports of Lemierre's syndrome (via PubMed search for "Lemierre's" in case reports), including our case, assessing initial symptoms and whether or not those patients visited a general practitioner in the outpatient setting and/or received any antibiotics (Table 1). We found that 9 out of 16 patients did see a healthcare provider (56%) with one of them having been previously hospitalized before being transferred to another facility. and 9 of those 10 patients received antibiotics (56%) [5–19].

Although our population size for this evaluation is small and it can be argued that these findings may not reflect an accurate picture of the patient population, Gulliford et. al reviewed cases utilizing the UK Clinical Practice Research Datalink and reached a similar conclusion, stating that while the incidence of peritonsillar abscesses and pneumonia may have increased from less aggressive antibiotic use, there was no evidence to support that same claim in the case for Lemierre's [20]. Therefore, we propose that future clinicians and researchers might benefit from evaluating other causes for the rising incidence of Lemierre's syndrome including, but not limited to, the possibility that known causative organisms are developing antibiotic resistance or that current detection methods have improved significantly, leading to greater detection rates.

The overall mortality of Lemierre's syndrome have decreased since its namesake clinical report but even in this modern age of antibiotics and more sensitive detection methods, the mortality can still be as high as 17% [1]. We propose that clinicians should be familiar with the signs and symptoms of potential cases (sore throat, neck mass, neck pain), have a low threshold for detection utilizing various modalities (neck ultrasound, CT chest, CT angiogram of the chest), and start aggressive antibiotic therapy as soon as possible [2].

Conflict of interest

We wish to confirm that there are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

Table 1
Comparison of initial symptoms and outpatient treatment in recent documented case reports.

Author (year)	Assessed as outpatient	Outpatient antibiotics prescribed?	Initial symptoms
Tawa et al. (2016) [3]	Yes	None	odynophagia, left cervical pain, and fever
Stefan et al. (2016) [4]	Yes	Clarithromycin	1-week history of severe sore throat and fever
Roland et al. (2016) [5]	Yes	Amoxicillin/clavulanic acid	dental infection present in the left inferior premolar for a month
Chamseddin et al. (2016) [6]	Yes	Amoxicillin	severe odynophagia and sore throat
Fielding et al. (2016) [7]	Yes	Clarithromycin	7-day history of worsening dysphagia, non-productive cough, hoarseness, fever, rigors, general malaise and anorexia
Panchavati et al (2017) [8]	Yes	Ampicillin/sulbactam and amoxicillin	fever, rigors and sore throat 7 days
Faraone et al. (2016) [9]	Yes	Amoxicillin/clavulanic acid	2-week history of sore throat, high fever, and mild neck tenderness
Kumral et al. (2017) [10]	No	None	7 day history of fever (Tmax 40.4C), left sided throat pain, fatigue, and myalgias
Budhram et al. (2017) [11]	No	None	1 week of right sided partial ophthalmoplegia and ptosis 4 month history of headache
Farhan et al. (2016) [12]	Yes	Unspecific IV antibiotics	two weeks history of right sided neck swelling associated with high grade fever, dysphagia, dysphonia and difficulty in opening her mouth
Birkner (2017) [13]	No	None	Previously fractured elbow Swelling of the left arm sore throat, shivering attacks, and fever accompanied by growing nausea
Meher-Homji et al. (2017) [14]	No	None	fever, pharyngitis and cervical lymphadenopathy
De Giorgi et al. (2017) [15]	Yes	Clarithromycin and ceftriaxone	occipital headache, malaise, hacking cough, chest pain exacerbated by inspiration, and fever for one month.
Kobayashi et al. (2017) [16]	No	None	Hemoptysis and a fever. pharyngeal pain
Osman et al. (2017) [17]	No	None	swelling of the right side of the neck (10 days) , fever, chills and difficulty swallowing (2 months)

We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscript has been approved by all of us.

We confirm that we have given due consideration to the protection of intellectual property associated with this work and that there are no impediments to publication, including the timing of publication, with respect to intellectual property. In so doing we confirm that we have followed the regulations of our institutions concerning intellectual property.

We understand that the Corresponding author is the sole contact for the Editorial process (including Editorial Manager and direct communications with the office). He/she is responsible for communicating with the other authors about progress, submissions of revisions and final approval of proofs. We confirm that we have provided a current, correct email address which is accessible by the Corresponding author and which has been configured to accept email from zhao.andrew@mayo.edu.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

References

[1] Kristensen LH, Prag J. Human necrobacillosis, with emphasis on Lemierre's syndrome. Clin Infect Dis 2000;31(2):524–32.
[2] Karkos PD, Asrani S, Karkos CD, Leong SC, Theochari EG, Alexopoulou TD, Assimakopoulos AD. Lemierre's syndrome: a systematic review. Laryngoscope 2009;119(8):1552–9.
[3] Riordan T. Lemierre's syndrome: more than a historical curiosa. Postgrad Med J 2004;80(944):328–34.
[4] Eilbert W, Nitin S. Lemierre's syndrome. Int J Emerg Med 2013;6(1):40.
[5] Tawa A, Raphaëlle L, Yannick M, Philippe S. Severe sepsis associated with Lemierre'S syndrome: a rare but life-threatening disease. Case Rep Crit Care 2016;2016:1–3.
[6] Stefan M, Klika D, Chrdle A, Smejkal P, Holub H. Lemierre's syndrome. Int J Infect Dis 2016;49:67.
[7] Roland T, Yombi JC, Yildiz H. Lemierre's syndrome presenting as acute thyroid Goiter. Vasc Med 2016;21(6):560–1.
[8] Chamseddin KH, Kirkwood ML. Lemierre's syndrome associated mycotic aneurysm of the external carotid artery with primary internal carotid artery occlusion in a previously healthy 18-year-old female. Ann Vasc Surg 2016;36:291.
[9] Fielding A, Pecheva M, Farghal A, Phillips R. Coexisting pulmonary haemorrhage and venous thrombosis: a tricky but novel case. BMJ Case Rep 2016. bcr2016217168.
[10] Panchavati PK, Kar B, Hassoun A, Centor RM. Fusobacterium necrophorum tonsillitis with mild case of lemierre's syndrome. Anaerobe 2017;43:102–4.
[11] Faraone A, Fortini A, Nenci G, Boccadori C, Mangani V, Oggioni R. Fusobacterium necrophorum pharyngitis complicated by Lemierre's syndrome. Case Rep Med 2016;2016:1–4.
[12] Kumral AV, Petersen Jr. WC, Heitz C, Waggoner-Fountain LA, Belyea BC. Lemierre'S Syndrome as a trigger for secondary hemophagocytic lymphohistiocytosis. J Pediatr Hematol Oncol 2017;39(6):e325–7.
[13] Budhram A, Shettar B, Lee DH, Silverman M, Gupta K. Bilateral cavernous sinus thrombosis in Lemierre's syndrome. Can J Neurol Sci 2017:1–3.
[14] Farhan A, Shah YA, Ali BT, Mumtaz U, Farooq U. The forgotten disease –Lemierre's syndrome. J Pak Med Assoc 2016;66(12):1652–5.
[15] Lukas Birkner. Lemierre's syndrome associated with Mechanical Ventilation and profound deafness. Case Rep Infect Dis 2017;2017:1–3.
[16] Meher-Homji Z, Mangalore RP, Johnson P DR, Chua K YL. Chromobacterium violaceum infection in chronic granulomatous disease: a case report and review of the literature. JMM Case Rep 2017;4(1).
[17] Giorgi A, Fabbian F, Molino C, Misurati E, Tiseo R, Parisi C, et al. Pulmonary embolism and internal jugular vein thrombosis as evocative clues of Lemierre'S syndrome: a case report and review of the literature. World J Clin Cases 2017;5(3):112.
[18] Kobayashi K, Hirotoshi M. Lemierre's syndrome with cavernous sinus thrombosis. Intern Med 2017;56(7):887–8.
[19] Osman M, Hasan S, Bachuwa G. Oesophageal cancer presenting as Lemierre's syndrome caused bystreptococcus anginosus. BMJ Case Rep 2017. bcr-2017-219661.
[20] Gulliford MC, Little P, Fox R, Charlton J. Safety of reduced antibiotic prescribing for self limiting respiratory tract infections in primary care: cohort study using electronic health records. BMJ 2016:i3410.