CASE REPORT

LEMIERRE SYNDROME: A FORGOTTEN INFECTION

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Lemierre's syndrome is a serious disease caused by an anaerobic bacteria called Fusobacterium necrophorum, which usually occurs in healthy teenagers and adolescents. The infection takes place in the throat and spreads through septic thrombosis of the tonsillar and internal jugular veins. The resulting bacterial infection is complicated by septic emboli in several places such as the lungs, joints and bones. Fever, pharyngitis, dysphagia, odynophagia, or swelling of the oropharyngeal is a common symptom. Although rare, there has been evidence of a recurrence of the condition in recent years, which may be associated with a reduction in the use of antibiotic therapy for the cold throat. The typical clinical image is characteristic, but many doctors do not recognize the condition and diagnoses are often delayed with potentially fatal consequences. We are presenting a similar case of Lemierre's syndrome in an elderly female with pharyngitis, resulting in thrombophlebitis, empyema and discharging neck sinus.

Keywords: Lumiere syndrome; Chest tube; Internal Jugular thrombosis

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INTRODUCTION

Andre Lemierre in 1936 described this condition in a patient of post-anginal septicaemia characterized by thrombophlebitis of the internal jugular vein and distant septic emboli.[1,2] The estimated incidence according to some studies is 3.6 cases per 1 million individuals per year. This low incidence is mostly because of the antibiotic use for treating oropharyngeal infections.[3] The majority of the cases are preceded by pharyngitis but it's not always the case. Some cases do occur after dental infection, intravenous catheter, tonsillitis and infectious mononucleosis.[4]

Lemierre's syndrome is a polymicrobial infection but Fusobacterium necrophorum is the most common organism responsible for Lemierre disease and is resistant to common antibiotics including macrolides, fluoroquinolones, tetracyclines, and aminoglycosides.[5] Other organisms include Eikenella, Porphyromonas, Streptococcus pyogenes, and Bacteroides. Beta-lactamase resistant beta-lactam antibiotics like co-amoxiclave, piperacillin-tazobactam, or meropenem are used empirically to treat this condition.[6]

CASE PRESENTATION

A 67-year-old female recently returned from Saudi Arabia (for Umrah) and presented with a history of sore throat one week back followed by fever, discharging anterior cervical sinus, cough, and shortness of breath. She had received some oral antibiotics with very little improvement. She was febrile but otherwise stable vitally with a blood pressure of 130/80 mm hg, the pulse of 95/ minute and respiratory rate of 20/minute. She was started on cefoperazone-sulbactam but no response was shown. Her TLC and CRP were raised. Her investigations are shown in the table.

Table-1: Investigations of the patient

Test	Day 1	Day 4	Day 6	Reference values
Hb	11.5 g/dl	11.0 g/dl	10.7 g/dl	12.5–16
TLC	16000/mm	12000/mm	7000/mm	4–11
CRP	25	12	1	<0.5 mg/dl
Na	138		136	135–150 mmol/l
k	4.4		4.8	3.5–5.5 mmol/l
cl	105		109	96–110 mmol/l
Creat	1	1.2		0.5–1.2mg/dl
Bilirubin	0.8			0.1–1 mg/dl
ALT	30			10–50mg/dl
Glucose ®	84			70–140 mg/dl

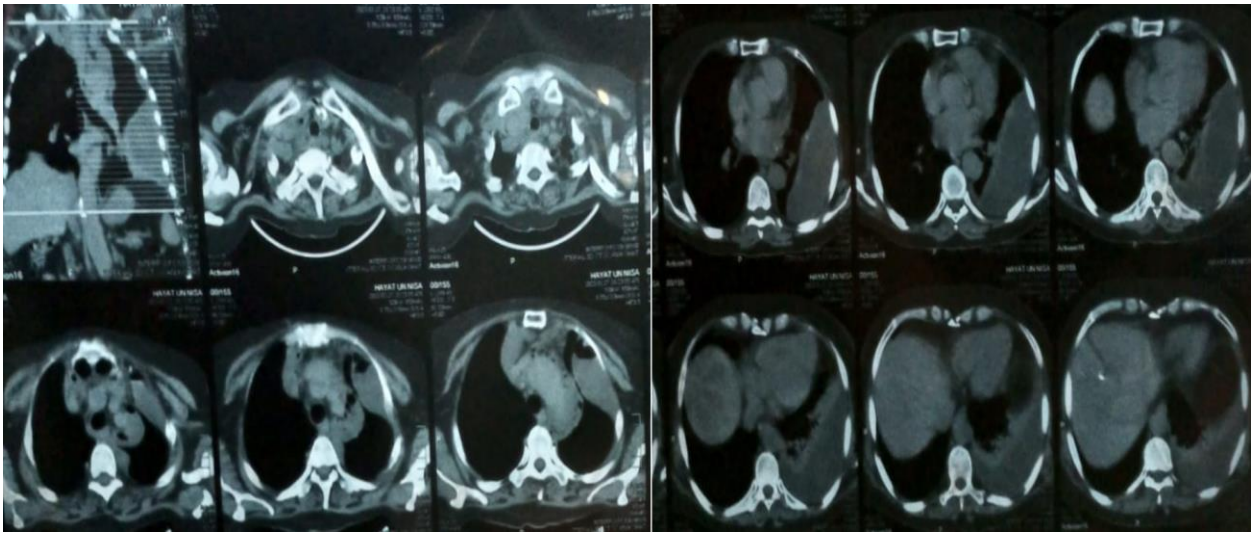

Figure-1: CT thorax images of the patient

X-ray chest showed left-sided intrapleural collection along with signs of pneumomediastinum which was confirmed by a CT scan of the thorax. A diagnostic pleural tape was done that showed puss and empyema was diagnosed. Subsequently, an ultrasound neck was performed for the draining sinus that showed collection on the right side of the neck with retrosternal extension, multiple air foci in the muscle plan and a filling defect in the right jugular vein.

A chest drain was passed on the left side and she was started on meropenem and clindamycin. Puss was sent for culture and sensitivity that showed the growth of normal flora. She was also started on anticoagulants but developed hemoptysis that led to discontinuation of the anticoagulants. Sinus drainage stopped on the 5th day and the patient was afebrile. On 6th day of admission, we managed to remove the chest drain and the patient was discharged on intravenous meropenem and clindamycin. On the 10th-day post-discharge follow-up, she was found to be afebrile, her sinus healed and she was mobile.

DISCUSSION

Oropharyngeal infection is common in society and the majority of the time is treated with oral or IV antibiotics**.** Because of the increased use of antibiotics**,** complications like Lemierre syndrome are rare and usually not under consideration.[7] The same was the case with our patient who was not suspected to cause complications. Although it is commonly found in young adolescents, our patient is an elderly lady with no co-morbid condition.[3] A prospective study in Denmark shows an annual incidence of 3-6 cases of Lemierre syndrome per million people aged 14-24 years.[8] It's common in men with a 2:1 ratio of male to female in contrast to our case which is an elderly female.

High-risk patients for developing complications regarding the type of infection are those with purulent tonsillitis or group A streptococcal pharyngitis where beta-lactam antibiotics should be used.[9] Macrolides and fluoroquinolones are frequently used for throat infections that can lead to resistance to medications as well as can lead to complications because of the increased resistance of fusobacterium to these medications.

Antibiotic stewardship is important for the prevention of antibiotic resistance as well as to avoid complications. A study in the UK by Gulliford et al demonstrated no increase in Lemierre syndrome with restricted use of antibiotics for acute respiratory tract infections (ARTI).[10] A large randomized study needs to be conducted to evaluate the association between the antibiotic stewardship program for ARTI and the incidence of Lemierre's syndrome.

Although there is no specific clinical manifestation that can tell us which patient will develop Lemierre syndrome high clinical suspicion, persistent high-grade fever and neck pain with tenderness in a patient with pharyngitis can be suspected to be at high risk of developing complications.[11] Clinical presentation can be different in different age groups with pharyngitis commonly in the young while distant complications like brain abscess and empyema in the elderly.

Fusobacterium is an anaerobic bacterium and the culture can take 6–8 days to grow which can lead to mismanagement of the disease. For this purpose, a diagnostic criterion has been suggested for Lemierre syndrome to avoid any delay in treatment commencement. This includes deep neck infections, subsequent septicaemia,

thrombophlebitis of the Internal Jugular Vein (IJV), and metastatic infections (ascending or descending septic emboli).[12] Necrotizing pneumonia is the common finding along with cavitary nodules suggestive of septic emboli. Other less common findings include pyomyositis, liver and spleen abscesses, endocarditis, pericarditis etc.

CT scan with contrast is the investigation of choice. It can detect IJV thrombosis, distant complications and brain abscesses.[13] Doppler ultrasonography can also be used to diagnose internal jugular vein thrombosis along with pleural effusion/empyema. A blood culture should be sent even if antibiotics are already used. MRI can be specifically used to detect IJV thrombus. In our case, we did a CT thorax and ultrasound neck that showed empyema and a filling defect in IJV suggestive of thrombus respectively.

A multidisciplinary approach should be adopted to manage Lemierre's disease. In collaboration with infectious disease experts, pharmacologists, radiologists, ENT specialists and thoracic surgeons, the disease can be diagnosed and managed effectively.[12] Appropriate antimicrobials along with surgical drainage are necessary. F. necrophorum is found to be sensitive to meropenem, clindamycin, metronidazole and co-amoxiclav.[13] We started our patient on meropenem and clindamycin along with chest drain and the patient responded well. She was followed up after 10 days of discharge from the hospital and declared cured.

CONCLUSION

Due to the rarity of the disease, clinical suspicion is very low for Lemierre's disease. Apart from usually found in young and adolescents, the elderly can also present with this disease therefore the threshold for the disease should be kept low. Any patient with pharyngitis not responding to antibiotics should be evaluated for this condition. CT thorax and neck with contrast plays an important role and should not be delayed.

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