Lemierre’s Syndrome caused by Klebsiella Pneumoniae: a case report

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ABSTRACT

Lemierre’s syndrome is a rare condition. Its manifestations are varied not only in clinical presentation but also in the infecting organisms. The most common organism causing this disease is Fusobacterium necrophorum. However, based on different underlying disease of the patient, other organism should be took into consideration to avoid delay antibiotics treatment.

A 53-years old male had poor controlled type 2 diabetes mellitus (DM). He came to our outpatient department with a palpable right neck mass for one week. Mild tenderness of the mass and dysphagia were also complained. Besides, body weight loss about 10kg in recent 1 month was noted. There was no fever, upper respiratory symptoms, or ear fullness. Initial impression was neck lymphadenopathy with unknown primary cancer. Chest computed tomography (CT) showed a 1cm cavitory nodule at left upper lung and a 4.7 cm mass lesion with central necrosis at right lower neck. The clinician suspected primary lung cancer with metastatic right neck lymphadenopathy. Surgical resection of left upper lung nodule for patholology and sampling right neck mass for cytology and culture were arranged. However, the pathologic report showed the lung nodule was organizing pneumonia and the right neck mass was only an abscess formation. Pus culture from right neck mass showed Klebsiella pneumonia (KP) growth. According to all of the examinations, the final diagnosis revealed the right neck mass was septic thrombophlebitis of internal jugular vein (Lemierre’s syndrome) caused by KP with pulmonary involvement.

Only three cases of Lemierre’s syndrome caused by KP in patients with DM had been reported previously. The association between DM and deep neck infection caused by KP is suggested but the mechanism is still unclear. Further statistics and pathogenesis study is needed. This paper reminds of this rare disease and its image findings for clinicians and radiologists to be aware of possible misdiagnosis and unnecessary surgical procedure. Otherwise, organisms other than F. necrophorum such as KP should be took into consideration especially in patient with DM.
in a patient with type 2 DM. The relevant studies are also reviewed.

CASE REPORT

In October 2014, a 53-years-old man with underlying disease of poor controlled type 2 DM came to our outpatient department with a right neck mass for one week.

Mild tenderness of the mass and dysphagia were complained. Poor appetite and body weight loss about 10kg in recent 1 month were also noted. There was no fever, upper respiratory symptoms, dental problems or ear fullness. Naso-oro-hypopharynx endoscope was done and there was no tumor. The initial impression of the right neck mass by clinician was lymphadenopathy with inflammatory process, and the other consideration is a metastatic lesion from esophageal or lung cancer. Thus, the patient was arranged hospitalization for primary cancer survey.

Panendoscopy and abdominal sonography revealed no tumor like lesion. Bronchofiberscopy showed no obvious endobronchial lesion and subsequent brushing and bronchoalveolar lavage showed negative finding for malignant cells. Serum tumor markers such as CEA, CA199, SCC, Free-T4, AFP, PSA were all negative. Other laboratory investigations revealed white blood cell count of 17.1 $\times$ 10$^9$ cell/L, (neutrophils 84.0%), hemoglobin 14.4 g/dL, platelet count 344 $\times$ 10$^9$ cells/L, international normalized ratio (INR) of 1.04, total cholesterol 203 mg/dL, triglyceride 160 mg/dL, alanine aminotransferase (ALT) 17 IU/L, aspartate aminotransferase (AST) 12 IU/L, uric acid 4.2 mg/dL, blood urea nitrogen 13 mg/dL, creatinine 1.0 mg/dL, total bilirubin 1.0 mg/dL and direct bilirubin 0.1 mg/dL during admission. Serum AC glucose was 297 mg/dL and HbA1c was 13.7% indicating poor diabetes control. Right neck mass content culture and sensitivity testing yielded KP growth; however, blood culture results were negative.

The chest CT showed a 1cm cavitory nodule with spiculated margin located at anterior segment of left upper lung (Fig. 1). Otherwise, there was no other pulmonary parenchymal lesion, no pleural effusion, nor mediastinal or hilar lymphadenopathy. A 4.7cm lobulated abscess formation with irregular peripheral enhancement in the right carotid space extending from the third to the fifth cervical vertebrae (Fig. 2) was seen. The right sternocleidomastoid muscle was swollen with surrounding inflammatory changes as well as

![Figure 1](image_url)

Figure 1. CT scan of the chest in lung window setting showing a cavitory nodule at anterior segment of LUL. The pathologic report is organizing pneumonia.

![Figure 2](image_url)

Figure 2. CT scan of the neck showing the abscess formation (Circle of arrowheads in 2a.) and thrombosis in the right internal jugular vein (white arrow). The black arrow is normal left internal jugular vein as opposed to right side.
Lemierre’s syndrome caused by Klebsiella pneumoniae

multiple enlarged cervical lymph nodes. The right internal jugular vein (white arrow) was thrombosed with only wall enhancement in the image. The thrombus also extended inferiorly down to the level close to right brachiophecalic vein. In opposition, the left internal jugular vein (black arrow) was intact with contrast medium passing through.

The clinician still highly suspected primary lung cancer with neck lymph node metastasis, so the wedge resection of the left upper lung nodule and intra-parenchymal lymph node were arranged. Besides, sampling right neck mass for cytology and culture were also done. The pathologic report showed that the left upper lung nodule was organizing pneumonia and there was no evidence of malignancy of intra-parenchymal lymph node. Pus culture from right neck mass showed KP growth. Base on above findings, the final diagnosis was right neck abscess formation due to septic thrombophlebitis of right internal jugular vein (Lemierre’s syndrome) with pulmonary metastatic organizing pneumonia caused by KP.

The patient underwent surgical incision and drainage of the abscess with adequate antibiotics treatment as well as blood sugar control. The clinical condition was improved and he was finally discharged after complete wound healing and the return of his biochemical parameters to normal levels.

DISCUSSION

LS has been reported to most commonly affect young, healthy adults, although cases in children have also been reported [8]. In a systematic review by Karkos, the most cases presented in the 2nd decade of life (51%), followed by the 3rd decade (20%) and then the 1st decade (8%). The most common first clinical presentation was sore throat, followed by a neck mass and neck pain. Prior three source of infection were tonsil, pharynx and chest. Otherwise, the prior three morbidity encountered in LS were brain, septic arthritis/osteomyelitis, and lung [4].

In radiographic features, identification of thrombophlebitis of the internal jugular vein is the first hard evidence to suggest Lemierre’s syndrome in many patients. In ultrasound (US), thrombus within the IJV may be noted but there is limitation of diagnostic accuracy under grayscale US. Imaging with Color Doppler US may overcome some of these shortcomings. However, the underlying site of infection is frequently not detected. Contrast enhanced CT(CECT) is a common choice of image study for the disease due to availability and its allowance for visualization of complications and underlying infection. In CECT, thrombosed veins would clearly be depicted due to its non-enhanced character with intraluminal filling defect. However, sometimes thrombosed IJV is mistook for neck abscess formation due to similar image appearance in axial view. The way to decrease misdiagnosis rate is to recheck the finding in coronal or sagittal view. If the lesion could be traced to subclavian vein, it should be a thrombosed IJV rather than an abscess formation. Otherwise, infection source such as tonsillitis, pharyngitis, or deep neck infection may be found on CT. Distant metastatic infection like pulmonary septic emboli or brain abscess is sometimes noted. Contrast enhanced MRI could have high grade confidence for diagnosis the same as CECT, but the availability and long examination time make it a second choice when the disease is in acute stage. Nevertheless, MRI plays an important role when the patient had poor renal function and could not receive contrast enhanced image. Conventional spin-echo sequences may demonstrate an absence of normal flow void in thrombosed IJV, and the signal change of the filling defect on T1 and T2 could help to differentiate septic thrombus from blood clot thrombus.

Fusobacterium species, most commonly Fusobacterium necrophorum, are responsible for the majority of bacteremia in cases of LS [3-9]. Some authors have questioned whether the isolation of F. necrophorum is really necessary for the diagnosis of LS [10]. A recent review reported 6% to 14% of cases of LS were associated with either negative cultures or organisms other than F. necrophorum [11]. The review study by Karkos P.D. [4] also showed only about 57% of the cases is caused by F. necrophorum. In our patient, there may be other possible reasons for negative blood cultures including prior antibiotic use or that F. necrophorum was overlooked, due to unfamiliar with its typical features and characteristics.

About pus culture from right neck mass of our patient revealed KP growth. So far, there are only three previously reported cases [3, 10, 12] of LS caused by KP. The common point of these cases and our patient is that they all have DM history and usually in poor blood sugar control status. Therefore, a question developed about whether DM has a role in selecting this organism, or it is a mere coincidence? Does poor blood sugar control predispose the patient to this organism? A recent review from Singapore [14] reported about 50% of their patients with DM developing deep neck abscesses and revealing KP isolated from the contents. A study from Taiwan reported by Huang et al. [13] showed that 98.4% of their patients with diabetes had infections due to KP, and suggested empirical antibiotics should cover KP in patients with deep neck infection who have DM.

CONCLUSION

Lemierre’s syndrome is a rare condition in antibiotics era, but it can be fatal with high mortality rate if untreated. The most common organism causing this disease is F. necrophorum, but other organisms should not be overlooked. Moreover, under specific condition such as the patient with diabetes mellitus, especially in poor control status, K. pneumoniae should be be taken into consideration as the source
of infection. Primary care providers have to be aware of this syndrome in patients with oropharyngeal infection who subsequently reveal symptoms and signs of neck mass accompanied with systemic illness like pulmonary involvement. Based on our case, if the final diagnosis could be made before, the patient should avoid unnecessary surgery. Radiologists also need to be familiar with the image pattern and make the accurate diagnosis to provide timely assistance for primary care providers.

REFERENCE

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