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A Case Report of an Unusual Presentation of Melioidosis

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Abstract

Melioidosis is a granulomatous infectious disease caused by Burkholderia pseudomallei. It is a motile, aerobic, oxidase-positive, gram-negative, non-spore forming soil saprophyte. It has gained a lot of importance in recent times as one of the most potent emerging infections in India. The clinical presentation of Melioidosis is highly variable, ranging from benign skin and soft tissue infections to rapidly fulminant and fatal septicaemia. It is because of this wide range of clinical diversity, it is called "Great mimicker" and poses diagnostic challenges to clinicians. In this case report, we illustrate the diagnostic difficulties we faced and our management of infrequent complication of disseminated Melioidosis presenting as broncho-oesophageal fistula with concomitant lung consolidation.

Keywords: Lung consolidation • Broncho- oesophageal fistula • Burkholderia pseudomallei • Melioidosis

INTRODUCTION

Melioidosis was first described way back in 1911 as "Glanders like" disease among morphine drug addicts in Burma, Rangoon by an Indian bacteriologist, C. S Krishnaswami with the help of a pathologist Whitmore. Burkholderia pseudomallei is present in the environment in a defined geographic distribution including much of South-east Asia and Northern Australia, where the infection is thought to be acquired after bacterial inoculation, ingestion or inhalation [1]. The first report of Melioidosis from India was by Raghavan et al., from Mumbai in 1991 [2]. Diabetes mellitus is the single most important risk factor associated with Melioidosis, other risk factors being thalassemia, renal disease, chronic lung disease, chronic alcoholism, and occupational exposure. It has bimodal age distribution and men are affected more often. The lung is the most common organ to be affected, presenting as an acute pulmonary illness characterized by prostration and marked toxicity which is usually out of proportion to physical findings or chest radiographic findings. It can also present as inapparent infections, transient bacteraemia, asymptomatic pulmonary infiltration, acute localized suppurative lesion, acute pulmonary infection, disseminated septicaemic infection, nondisseminated septicaemic infection or chronic suppurative infection and many other atypical forms.In this case report, we illustrate one such atypical and rarepresentationof this increasingly reported and important disease [3].

CASE REPORT

A 48-year-old male from Assam, a teacher by profession, presented with chief complaints of decreased appetite, difficulty in swallowing, and weight loss for last six months, fever for four months duration and cough with mild to moderate expectoration for four months. He was initially evaluated in a local hospital in Assam, and there he received IV antibiotics. But patient clinical condition continued to get worsened. On evaluation, he was drowsy, febrile and hypotensive. Bilateral crepitations were heard at the lung bases on auscultation. Routine haematological investigations revealed haemoglobin 8.3gm/dl, total leukocyte count of 5910 /mm3, Platelet count of 1.05lakh/cumm, elevatedESR (113 mm). Liver enzymes were elevated (Aspartate transaminase - 194 U/L, alanine transaminase - 168 U/L and alkaline phosphatase - 144 U/L). Serum sodium was 134 meq/lit and serum potassium was 3.2 meq/lit; Blood Urea – 21mg/dl and Creatinine - 0.5mg/dl. Random blood glucose was 44mg/dl. 25% Dextrose and double strength inotrope support were given in view of hypoglycemia and hypotension. Blood cultures were sent, and started him on empirical IV antibiotics (Inj. Meropenem 1gm IV q 8 hourly and Inj Doxycycline 100mg IV q 12 hourly). Review of old chest radiograph showed opacities in bilateral upper, right middle and left lower lobes.

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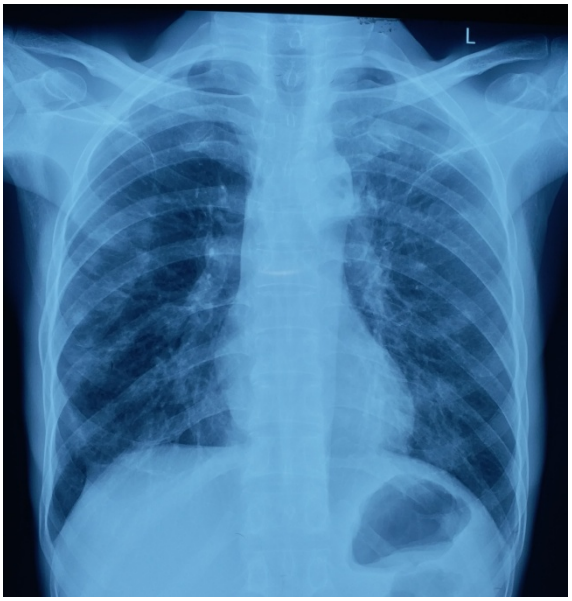
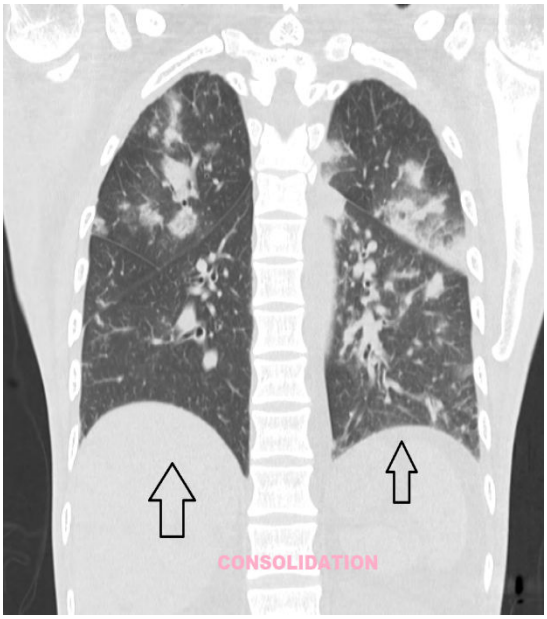


FIGURE 1: Chest X-ray showing areas of opacities in bilateral upper, right middle and left lower lobe.

Computed tomography (CT) scan chest showed areas of consolidation in both upper lobes, right middle lobe, lingula and left lower lobe, cavitations in two of the lesions in the left upper lobe, bilateral pleural effusion.



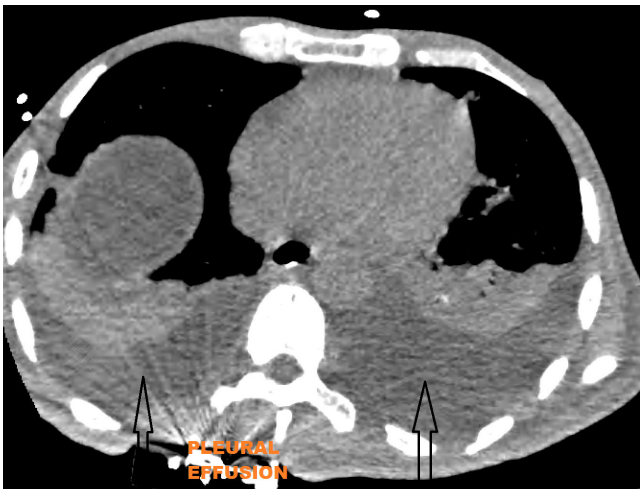


FIGURE 2& 3: CT- Chest showing areas of consolidation in bilateral upper, right middle, lingula and left lower lobe; bilateral pleural effusion.

Because of a patient complaining of difficulty in swallowing food, we did UGI endoscopy, and itshowed anoesophageal ulcer and possible tracheo-oesophageal fistula.

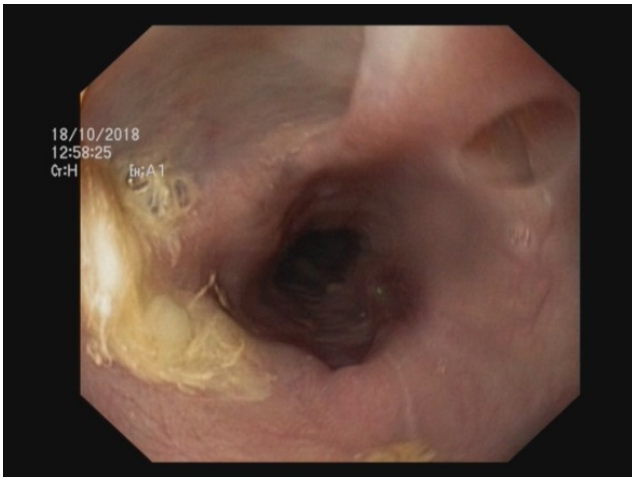


FIGURE 4: Upper GI endoscopy showing possible tracheo-oesophageal fistula

Bronchial wash was done, and samples sent for bacteriology tests, XPERT MTB and cytology and were non-contributory. Because of a suspected tracheo-oesophageal fistula, the patient underwent oral contrast computed tomography (CT) study. It showed evidence of oral contrast in the left main bronchus, suggestive of Left main broncho-oesophageal fistula.

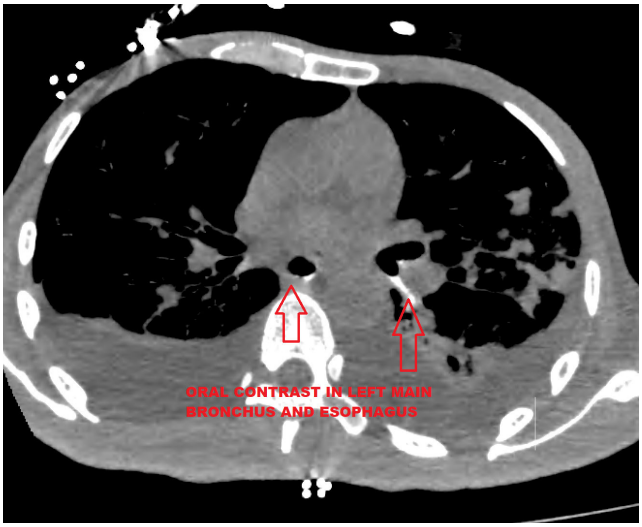


FIGURE 5: Oral contrast CT study showing contrast in left main bronchus and oesophagus

Blood cultures grew Burkholderia Pseudomallei. Bronchial wash also grew Burkholderia Pseudomallei. The sensitivity testing and interpretation were done by disk diffusion as per Clinical and Laboratory Standards Institute recommendations and found to be sensitive to cotrimoxazole, Ceftazidime. Meropenem and Doxycycline were stopped and started him on Inj. Ceftazidime 2gm IV, q 6th hourly, was also initiated on Bactrim DS oral twice daily.

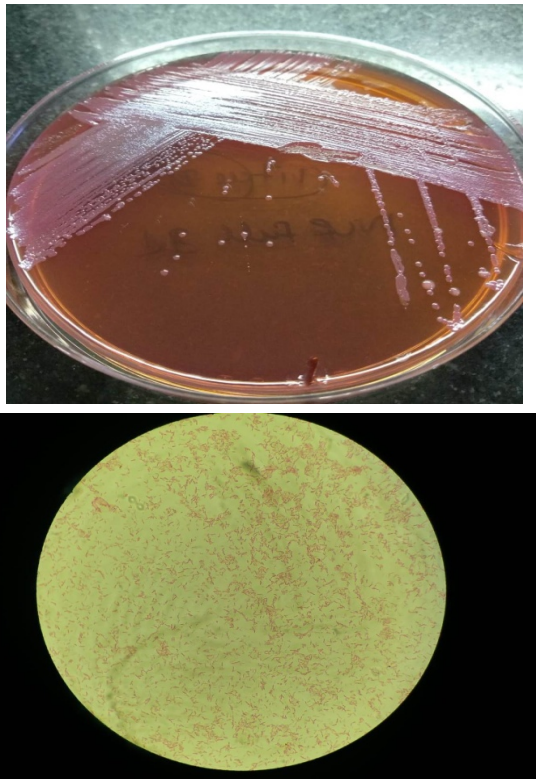


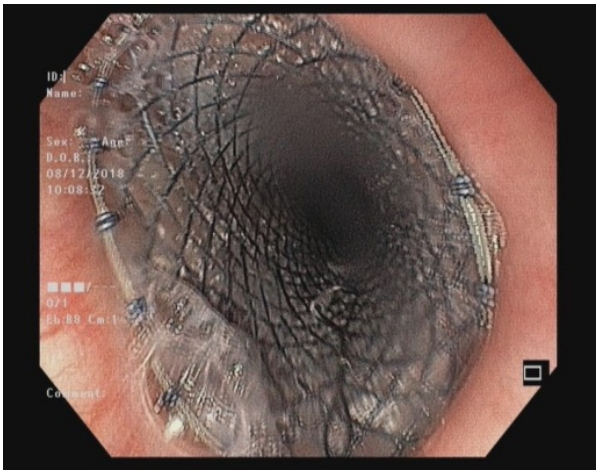
FIGURE 6: MacConkey agar with FIGURE 7: Gram-negative bacilli with

Non-fermenting, wrinkled colonies of bipolar staining (Safety pin appearance)

Burkholderia pseudomallei with metallic

Sheen

The final diagnosis was disseminated Melioidosis with left main broncho-oesophageal fistula.A multidisciplinary team of the primary physician, surgical gastroenterologist, medicalgastroenterologist and radiologist have discussed and arrived at a consensus that the best plan of treatment was to stent the fistula at the site of opening in the oesophagus. Hence he underwent oesophageal stent placement. It was planned to leave the stent in position for one month and review the patient for extraction of the stent then.



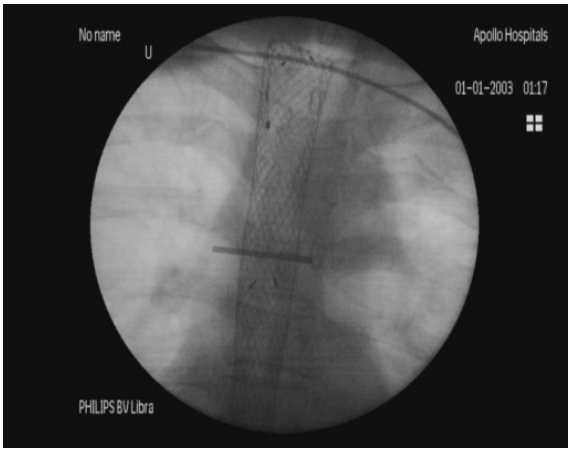


FIGURE 8&9: Esophageal stent in place at the site of the fistula

Patient general condition improved during the hospital stay. Reasonable glycemic control was achieved, and he had stable haemodynamics. He was discharged with advice to continue oral cotrimoxazole for three months.

DISCUSSION

Melioidosis which was previously considered to be a rare infection is emerging rapidly in the Indian subcontinent. The exact reasons for the increased incidence of Melioidosis in India in recent years remain unclear. The increased detection is due to considering the diagnosis in all patients with severe sepsis/abscess/ septic arthritis /severe pneumonia and routinely speciating all non-fermenters grown in culture. Besides, diabetes is increasing in the Indian population and may be driving an increase in the incidence of the disease [2]. Most cases are reported from Northeastern India and West Bengal, though there have been case reports from the southern part of India also. Our patient is from the northeastern part of India. Diabetes mellitus is the single most important risk factor associated with Melioidosis. A study was done on Melioidosis at our centre by Dr R Gopalakrishnan et al. between 2005 and 2010 showed diabetes as most commonly associated risk factor followed by alcoholism.

Underlying risk factor	
Diabetes mellitus	14(43.75%)
Alcohol consumption	7(21.87%)
Trauma	2(6.25%)
Pregnancy	1(3.12%)

SOURCE: R. Gopalakrishnan et al. “Melioidosis: An Emerging infection in India” Journal of the association of physicians of India, September 2013, Vol. 61

In our case, he was a known diabetic which was poorly controlled. Melioidosis is rightly called as "Great mimicker" in view of its wide range of clinical diversity. The disease can manifest as transient bacteraemia, asymptomatic pulmonary infiltration, acute localized suppurative lesion, acute pulmonary infection, disseminated septicaemic infection, nondisseminated septicaemic infection or chronic suppurative infection and many other atypical forms. Because of this wide clinical diversity melioidosis should be suspected in any severely ill febrile patient with an underlying predisposing condition who lives in, or has travelled from an endemic area [4]. The most common for Melioidosis seen clinically is community-acquired pneumonia. However, many of these patients present with prolonged fever, weight loss, and suspicious chest X-ray findings, are misdiagnosed as tuberculosis and may get anti-tubercular therapy.

Though there have been many case reports of severe pulmonary and GI involvement in Melioidosis, we did not get any case report of Melioidosis causing Broncho- oesophageal fistula even after searching the world literature extensively. In the background of poorly controlled diabetes mellitus, with multiple issues like electrolyte disturbances, hypoglycemia, hypotension, inadequate oral intake, it was quite challenging in managing the case. Our primary goal was to stabilize the patient hemodynamically before we subject him to any invasive investigation. Once he was stabilized, Computed tomography (CT) scan chest, upper GI endoscopy and Bronchoscopy were done. Meanwhile, started him on empirical IV antibiotics therapy, and his electrolytes, blood sugars and blood pressure were monitored regularly and managed appropriately. Once culture reports confirmed Melioidosis, he was immediately started on Ceftazidime and continued for two weeks. It was more challenging for the closure of the fistula between left main bronchus and oesophagus. A multidisciplinary team including Surgical gastroenterologist, Medical gastroenterologist, cardiothoracic surgeon were all involved, and it was decided that endoscopic stenting and closure of fistula opening in the oesophagus is the best approach and same was done successfully. The patient responded well to IV ceftazidime therapy. He was discharged in a hemodynamically stable condition and advised to continue cotrimoxazole for at least four months.

Our case highlights the importance of considering Melioidosis as one of differential diagnosis in any patient presenting with prolonged duration of fever and any atypical form of presentation in the background of diabetes and also the challenges we may come across while managing these cases with out-of-the-box presentations. Possibly this is the first case report of Melioidosis presenting with Broncho- oesophageal fistula and concomitant lung consolidation.

CONCLUSION

Being one of the most potent emerging infections in India, the clinicians and microbiologists should be suspicious of B. pseudomallei in any suppurative lesions at multiple sites [3]. Culture is the gold standard test for diagnosis of Melioidosis. All cases with severe septicaemic Melioidosis should be treated with parenteral antibiotic therapy. The current recommendations for antibiotic

treatment include an initial intravenous intensive phase for 10-14 days, followed by an oral eradication phase [5]. Ceftazidime/ carbapenems are the agents of choice for initial intensive phase and oral cotrimoxazole/ Doxycycline for eradication phase. There have been case reports of emerging ceftazidime resistant cases in India. SO, we should be very careful and cautious in choosing antibiotics. In ceftazidime resistant cases, Imipenem with Doxycycline combination therapy for the initial intensive phase followed by oral cotrimoxazole for eradication phase can be considered as an alternative regimen. Awareness amongst clinicians and microbiologists is vital to be able to diagnose this emerging infection and starting patient on appropriate antibiotic therapy at the earliest for better outcomes. This case highlights the possibility of Melioidosis in cases with broncho-oesophageal fistula with lung consolidation and septicemia in the background of diabetes.

References

1. Gopalakrishnan, R "Melioidosis: An Emerging Infection in India." *Journal of the association of physicians of India.* (2013): 61

2. Karuna, Tadepalli Sagar Khadanga , Dharmendra Dugar , Biyanka Sau and Priyadarshini Bhoi “Melioidosis as a Cause of Acute

3. Puthucheary, Savithri Devi. "Melioidosis in Malaysia." *Medical Journal of Malaysia.* 64 (2009): 266-274.

4. Abdomen in Immuno-Competent Male from Eastern India.” *J Lab Physicians.* 7 (2015) :58-60

5. Jayaram, M. "Melioidosis-Should it be a Notifiable Disease in Malaysia?." *Medical Journal of Malaysia.* 60 (2005): 531.

6. Li, Philip H, Chi Hung Chau, and Poon Chuen Wong. "Melioidosis Mycotic Aneurysm: An Uncommon Complication of an Uncommon Disease." *Respir Med Case Rep.* 14 (2015): 43-46.

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