Recurrent Melioidotic Neck Abscess

V Sha Kri Eh Dam[1]

ABSTRACT

Background

Melioidotic neck abscess is a rare complication of chronic infectious disease caused by *Burkholderia pseudomallei*. Recurrent cases with laboratory confirmation, which could be caused by incomplete eradication of infection or immunocompromised state of patient, have never been described in the literature. Its management is challenging, with no consensus to date of treatment regime.

Case presentation

We present a case of a young patient with multiple comorbidities presenting with recurrent melioidotic neck abscess, successfully treated with surgical drainage and a long-term combination of antibiotics.

Conclusion

There are wide range clinical manifestations of melioidosis with pneumonia is the most common presentation, while neck abscess is rarely reported. The bacteria may remain inactive for some time and re-activate when the host immune system has deteriorated. Fail to completely eradicate the disease may predisposes to another episode of complication. Longer duration of treatment and follow-up should be considered, especially in immunocompromised patients, and the patients should be managed under multidisciplinary teams to minimize the risk of recurrent and catastrophic complications.

Keywords

recurrent neck abscess, melioidosis, Burkholderia pseudomallei

INTRODUCTION

Neck abscess is commonly seen in immunocompromised patients especially diabetes mellitus (DM). Odontogenic and upper respiratory tract infections are among the most common source of the abscess formation[1,2]. The majority of odontogenic infection is secondary to dental caries[3]. Asian countries have different microbiology profile compared to Western countries[1]. Gram-negative bacteria, particularly *Klebsiella pneumonia*, are predominant organisms in Asia and have a strong relation with DM[1,2].

Burkholderia pseudomallei is another important gram-negative organism but rarely reported in neck abscess cases. Its infection is also known as melioidosis and has extremely varied clinical manifestations, ranging from localised infection to multiorgan involvement and fulminant septic shock[4]. To the best of our knowledge, recurrent neck abscess secondary to melioidosis with laboratory confirmation has never been described in the literature. The management is challenging and different from other more common organisms.

CASE PRESENTATION

A 28-year-old lady presented with left neck swelling for one week duration. It was progressively increasing in size and was associated with pain on movement. There was no history of fever, upper respiratory tract or dental infections, and no symptoms of upper aerodigestive tract obstruction. She had multiple medical problems, namely, DM, haemoglobin H disease with blood

Correspondence

V Sha Kri Eh Dam, Department of Otorhinolaryngology-Head & Neck Surgery, Hospital Lahad Datu, Peti Surat 60065, 91110 Lahad Datu, Sabah, Malaysia, E-mail: kridamrong@gmail.com

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transfusion dependence, and hepatitis C. In addition, she had history of right neck abscess (Figure 1) complicated with right internal jugular vein (IJV) thrombosis (Figure 2) one year earlier, which required incision and drainage (I&D) under general anaesthesia. Pus sent during the operation for culture and sensitivity had shown growth of *Burkholderia pseudomallei*; thus, the diagnosis of melioidosis was made. She had been referred to the infectious disease team and started on intravenous ceftazidime for two weeks, followed by a long-term oral amoxicillin clavulanate for 3 months based on the bacteria culture sensitivity. She was also started on long-term oral antiplatelet (Cardiprin) for her IJV thrombosis. On examination, there was a neck swelling at the left posterior triangle, measuring 4 cm x 4 cm in size, inflamed overlying skin with presence of a punctum and firm and tender on palpation (Figure 3). The previous I&D scar on the right side of the neck was well healed. An ultrasound scan of the neck was performed and showed a well-defined heterogeneous hypoechoic collection at the left posterior triangle, measuring 2.3 cm x 3.6 cm x 3.9 cm (Figure 4), associated with multiple adjacent cervical lymphadenopathies. She was subjected for I&D under general anaesthesia. The pus for culture and sensitivity again showed the growth of *Burkholderia pseudomallei*. In light of the culture sensitivity, she was started on intravenous ceftazidime for four weeks, followed by oral amoxicillin clavulanate and doxycycline for 6 months. Ultrasound abdomen was also performed to look for associated intrabdominal abscesses but showed normal findings. The neck wound healed well after 6 weeks with daily wound dressing. The patient was managed under multidisciplinary teams, including haematology, endocrinology, and infectious disease, to optimise her medical and immunocompromised conditions with the aim of preventing a recurrence of the abscess and other complications. There was no recurrence after 15 months of follow-up.

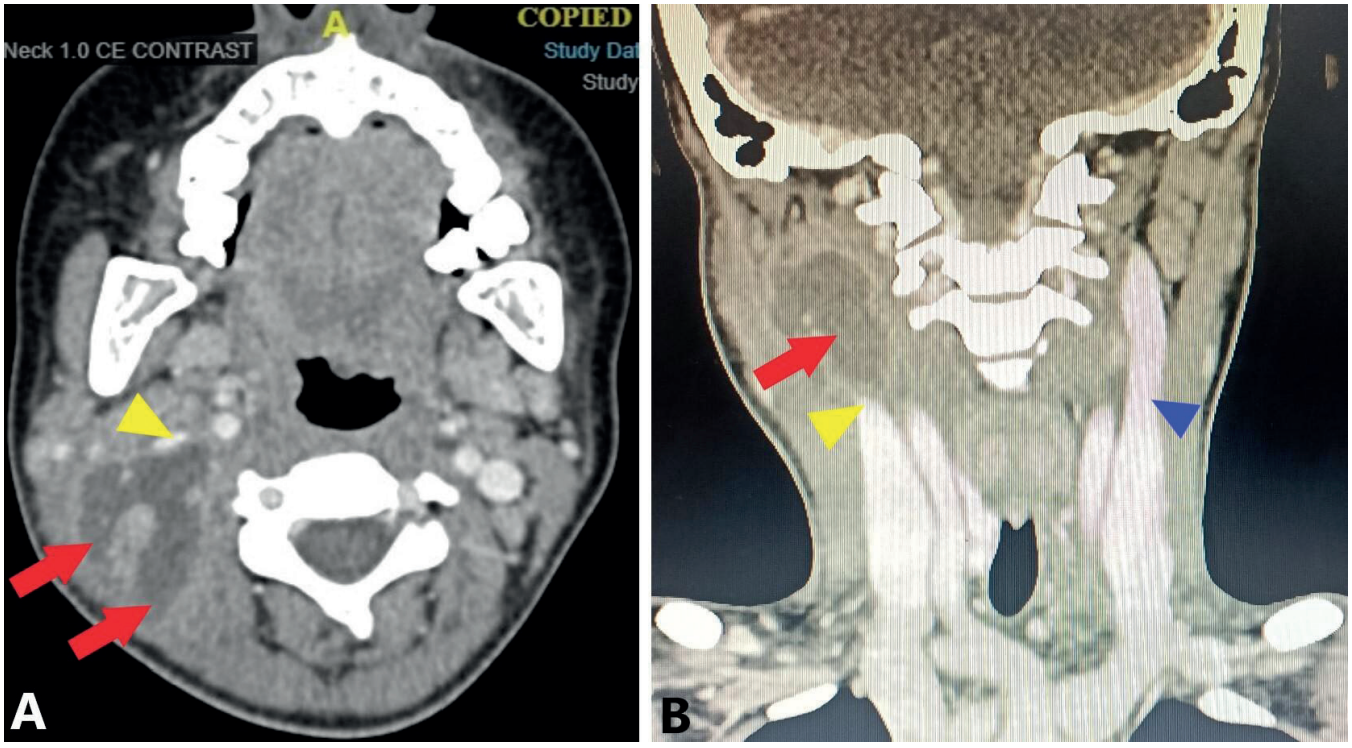

Figure 1. Contrast enhanced computerized tomography of neck in axial (A) and coronal (B) views shows a peripheral rim enhancing collection (red arrow) at the right posterior cervical space involving the right peri-vertebral and paraspinal spaces, measuring 3.2 cm x 2.5 cm x 4.0 cm, with compression of right internal jugular vein (yellow arrowhead). Note the left internal jugular vein is pattern (blue arrowhead).

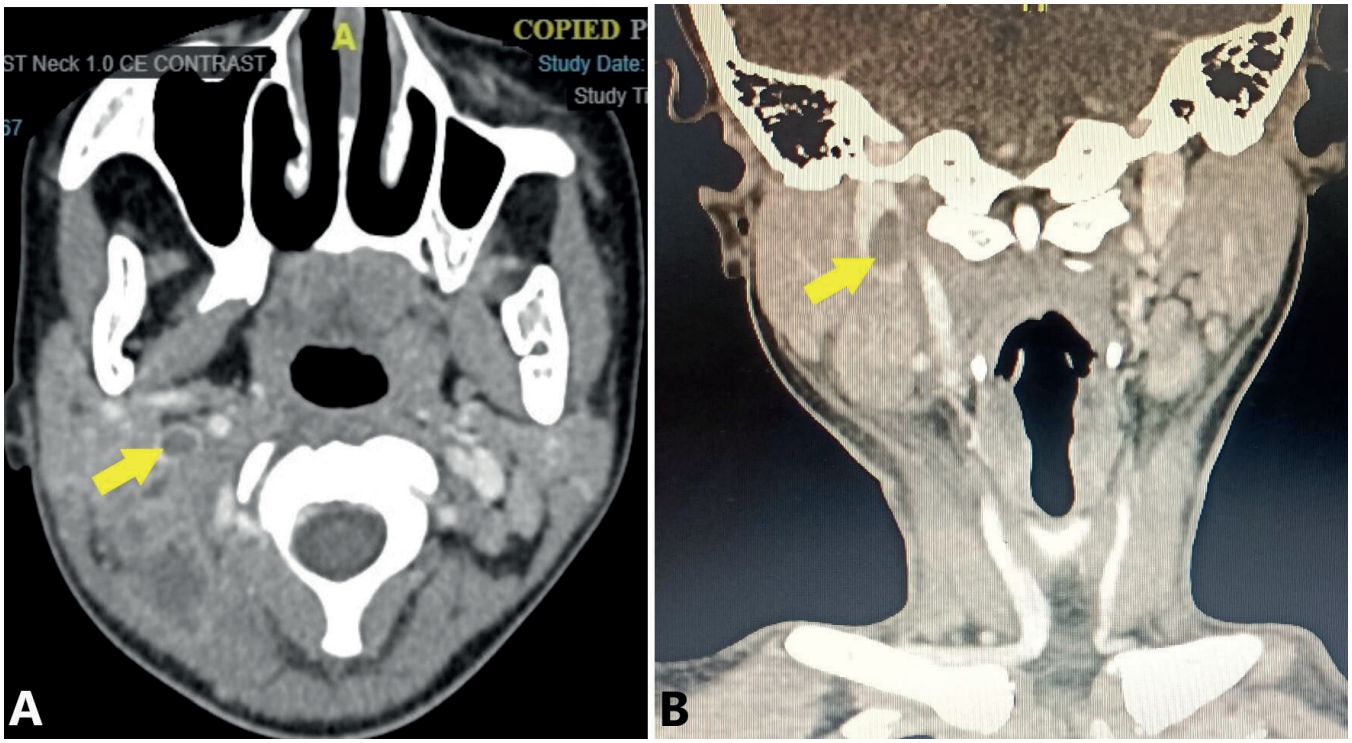

Figure 2. Contrast enhanced computerized tomography of neck in axial (A) and coronal (B) views shows filling defect (arrow) of the right internal jugular vein, measuring 1.5 cm in length, in keeping with thrombosis.

Figure 3. A 4 cm x 4 cm neck swelling at the left posterior triangle, inflamed overlying skin with the presence of a punctum.

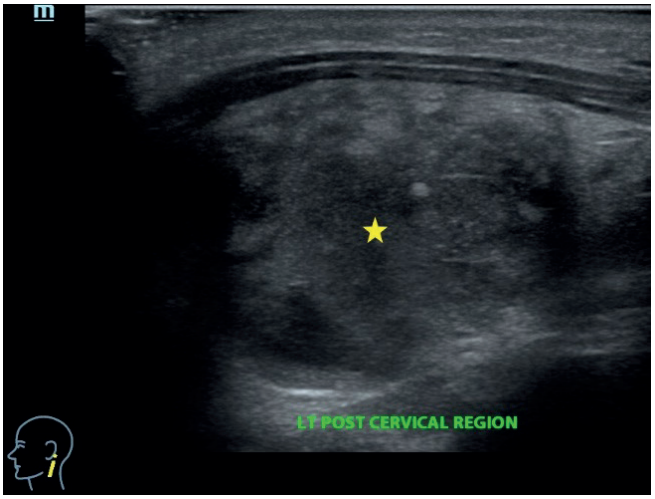

Figure 4. Ultrasound scan of the neck shows a well-defined heterogeneous hypoechoic collection at the left posterior triangle, measuring 2.3 cm x 3.6 cm x 3.9 cm (centre of collection marked with yellow star).

DISCUSSION

Recurrent neck abscess is rare and usually associated with congenital anomalies like thyroglossal duct cyst, branchial cleft anomalies, and lymphangioma[5]. The

absence of these anomalies should raise the suspicion of host immunological disorder or atypical causative pathogen, as demonstrated in the present case.

Melioidosis is more common in Southeast Asia and Northern Australia and rare in Western countries[6]. It is caused by *Burkholderia pseudomallei* infection, which is a gram-negative aerobic bacillus organism. This bacterium is mostly inhabitant in the soil and water of endemic tropical and subtropical regions and mostly affects farmers and those from rural areas[6]. In Western countries like Europe and the United States, patients are usually immigrants or have a history of travelling to endemic areas[4,6]. Apart from skin inoculation, which is the main route of infection, current evidence also suggests that inhalation of pathogens and ingested contaminated water can be other paths of infection[7].

Pneumonia is the most common manifestation of melioidosis, followed by genitourinary and skin infections and formation of abscesses in internal organs, especially the liver and spleen[4]. Ultrasound abdomen is routinely performed in our centre to look for any associated intrabdominal abscess. Other less common presentations are neck abscess, parotitis, lymphadenitis, mycotic aneurysm, mediastinal mass, pericardial effusion and pancreatitis[8]. Due to wide clinical manifestations, the mortality rates were reported as high as 95% in untreated and 50% in treated patients[4].

In the present case, we are not sure about the route of infection, but the patient has significant risk factors for infection, namely, DM, haemoglobin H disease and hepatitis C. In addition, the patient is transfusion dependent due to the haemoglobin H disease, which may have a higher risk of transfusion-transmitted infection. The incidence of transfusion-transmitted melioidosis is rarely reported, but there are reported cases of bacteraemia melioidosis in 74% of thalassemia patients in East Malaysia, with a mortality rate as high as 50%[9].

Recurrent melioidotic neck abscess with laboratory confirmation has not been reported previously, even in endemic areas like Malaysia. Selladurai et al. had reported a case of recurrent neck abscess in melioidosis patient but was treated empirically with antituberculosis at initial presentation due to first pus culture showed no growth[10]. In contrast, our presenting case had confirmed growth of *Burkholderia pseudomallei* during the first and second presentations. The recurrence could be due to relapse or reinfection. Relapse is the most common scenario, but reinfection should be considered if the recurrence occurred after two years[11]. In this presenting case, we believed the recurrence are due to the immunocompromised state of the patient and incomplete eradication of the infection. Melioidosis is a chronic infection and may behave like tuberculosis to a certain extent. The bacteria may remain inactive for some time but have the capability to re-activate when the host immune system has deteriorated. The severity of disease, choice and duration of antibiotic, and compliance to medication may explained the failure of complete eradication in some patients. Therefore, apart from acute management like I&D of the neck abscess, long-term management with appropriate antibiotics is crucial to properly eradicate the infection.

United States Centres for Disease Control and Prevention guidelines recommended ceftazidime and meropenem as the first-line antibiotics during intensive phase of treatment[8]. The duration of treatment is at least two weeks, or longer in critically ill patients, deep-seated collections or organ abscesses, extensive pulmonary disease, septic arthritis, osteomyelitis, and neurologic melioidosis. During eradication phase, amoxicillin clavulanate, trimethoprim sulfamethoxazole and doxycycline are commonly used antibiotics with a total duration of 3 to 6 months. Higher recurrence rates were seen in patients treated with amoxicillin clavulanate, ciprofloxacin-azithromycin, doxycycline monotherapy and quinolone monotherapy[11]. There was no consensus on the duration and treatment regime of melioidotic neck abscess, but we believe a longer duration and combination of antibiotics based on sensitivity should be considered, as seen in the present case. The patient should be managed by multidisciplinary teams to improve the immunocompromised state and prevent recurrent and other complications.

CONCLUSION

Melioidotic neck abscess is a rare infectious disease with no consensus on duration and treatment regime. Longer duration of treatment and follow-up should be considered, especially in immunocompromised patients, to prevent recurrent and catastrophic complications. A multidisciplinary team approach is always ideal in managing this chronic infection.

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Conflict of Interest

The author has disclosed no conflict of interest.

Ethical Approval Issue

There was no ethical approval sought other than getting consent from the patient.

Authors' contribution

Data gathering, conception, design, and critical revision of the article for important intellectual content: V Sha Kri Eh Dam

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