



A Rare Case of Complicated Sinonasal Melioidosis Mimicking Sinonasal Lymphoproliferative Disease

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Abstract Unilateral nasal obstruction with purulent nasal discharge is one of the presentations for sinonasal melioidosis. However, it may mimic nasal NK/T cell lymphoma. Both causing tissue destruction involving nasal septum, lateral nasal wall and palate. Here, we report a case of disseminated melioidosis involving sinonasal mimicking nasal lymphoma in a 32-year-old immunocompetent lady. She presented with prolonged fever, unilateral nasal blockage, painful facial swelling and knee pain. Clinical findings revealed extensive necrotic tissue and crusting involving right lateral wall of nasal cavity. Tissue and blood culture and sensitivity (C + S) grew *Burkholderia pseudomallei*. Recovery was complete after surgery and antibiotics.

Keywords Rhino-sinusitis complications · Endoscopic sinus surgery · Infectious disease · Bacteriology · Rhinitis

Introduction

Melioidosis, also known as Whitmore disease is a tropical bacterial infectious disease caused by *Burkholderia pseudomallei* [1]. It is endemic in South-East Asia and Northern

Australia [2]. It carries overall mortality rate of 20.5% [3]. The disease has diverse clinical manifestations including pneumonia, genitourinary infection, soft tissue infections, and joint infections [3, 4]. About 25–30% of melioidosis cases in Malaysia present as disseminated disease [4, 5]. Patients may acquire the disease via inhalation or hematogenous spread following inoculation of the pathogen [4, 5]. Sinonasal disease could be a focus of infection in melioidosis and causing local and systemic spread of the disease [6]. Meanwhile, sinonasal lymphoma is a rare sinonasal solid tumour which commonly affect maxillary and ethmoid sinus [7–9]. The most prevalent histological type is diffuse large B-cell lymphoma, followed by extranodal natural killer/T-cell lymphoma [8, 9].

Case Report

A 32-year-old female of Iban heritage was referred to Otorhinolaryngology Head and Neck Surgery (ORLHNS) Department with the complaint of painful right facial swelling, fever, malaise, and right knee pain for one week. It was preceded by one month history of progressive right nasal obstruction, foul smelling discharge and intermittent fever. She has a medical history of hypertension and on a single antihypertensive agent. Otherwise, she was non-diabetic and not in an immunocompromised status. Prior to her presentation, she sought treatment at multiple general practitioners, however had no resolution of her symptoms despite multiple courses of antibiotics Fig. 1.

On examination, the patient appeared septic. She was febrile and tachycardic (heart rate: 138 beats per minute). There was right facial cellulitis involving the right periorbital and maxillary region. Visual acuity and eye movements were normal. Naso-endoscopic examination

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Fig. 1 Cellulitis involving right facial and periorbital region with right nasal discharge

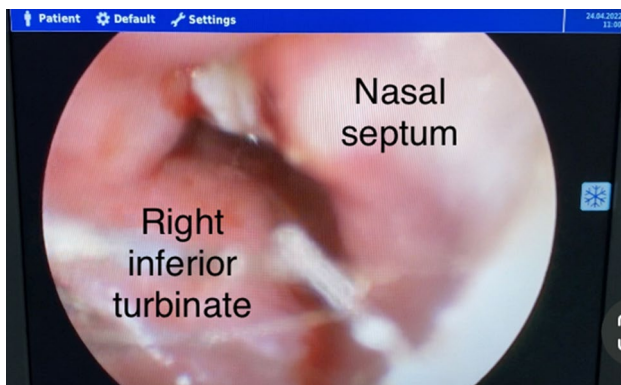


Fig. 2 Endoscopic view of right nasal cavity with erythema and oedema of the mucosa

revealed a very oedematous and erythematous nasal cavity mucosa with friable mucosa tissue, crusting, necrotic patches at right middle meatus and purulent nasal discharge Fig. 2.

Furthermore, she also had a left knee swelling associated with pain and difficulty in weight bearing. She was referred to our orthopaedic service for further evaluation and management. Fig. 3.

Blood investigations revealed normal total white cell count but an elevated C-reactive protein level of 345.2. Other blood parameters such as renal function, blood glucose level were normal. Screening tests for HIV, syphilis, hepatitis B and C were negative, indicating that the patient did not have an underlying immunocompromised status Fig. 4.

Computed tomography of paranasal sinus (CT PNS) showed soft tissue density mass filling the right maxillary and ethmoid sinuses. There were features of invasive sinusitis, cheek rim-enhancing collection, resulting in paranasal sinus obstruction, associated with right preseptal cellulitis Fig. 5.



Fig. 3 Axial view of CT PNS showed soft tissue density within right ethmoidal sinus



Fig. 4 Axial view of CT PNS showed soft tissue density within right nasal cavity and right maxillary sinus with medial wall of maxillary sinus thinned out

Patient was admitted and started on intravenous (IV) Ceftriaxone 2 g OD and IV Metronidazole 500 mg thrice daily (TDS). Emergency right endoscopic endonasal sinus surgery was performed. Necrotic tissue was debrided. Limited right medial maxillectomy and ethmoidectomy were done. Intra-operatively there was necrotic and inflamed mucosa



Fig. 5 Resolved right facial and periorbital cellulitis

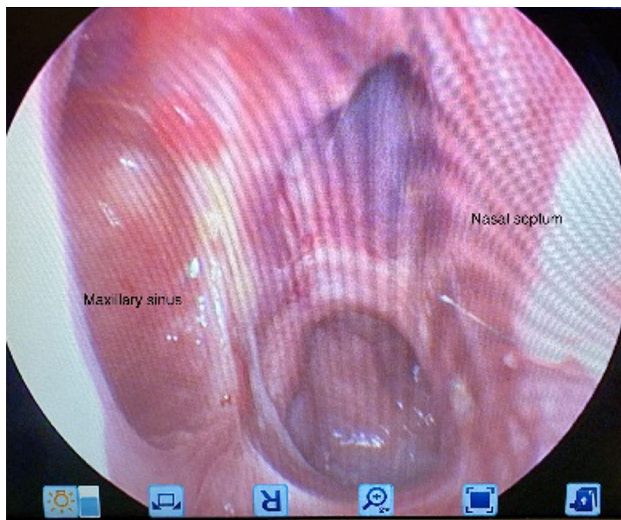


Fig. 6 Clear maxillary sinus

involving entire right nasal cavity, right maxillary, anterior and posterior ethmoid sinuses. Multiple areas of right nasal cavity tissue were taken for histopathological examination (HPE) and culture and sensitivity (C&S) were sent Fig. 6.

Knee joint arthroscopy and washout were done. Intra-operative findings showed inflamed synovium and turbid synovial fluid with abundant debris. Upon discharge, there was no further follow-up by orthopaedic team.

HPE showed extensive necrotic tissue. Tissue and blood C&S grew *Burkholderia pseudomallei*, sensitive to Amoxicillin/Clavulanic acid, Ceftazidime, Imipenem, Sulfamethoxazole-trimethoprim. The diagnosis of sepsis secondary to disseminated melioidosis was made and the antibiotics were escalated to IV Meropenem. The patient stayed in intensive care unit (ICU) for 12 days due to metabolic acidosis secondary to sepsis and then subsequently three weeks in the general ward. The patient completed one week of IV Meropenem 1 g thrice daily (TDS) and three weeks of IV

Ceftazidime 2 g four times daily (QID) in ward and changed to oral Bactrim four tablets twice daily (BD) upon discharge. She also received nasal douching four to six hourly, regular weekly nasal toileting and topical nasal decongestants.

During follow up at three months post operation, the patient has completed three months of oral Bactrim and completely recovered, with no residual symptoms of the disease.

Discussion

Burkholderia pseudomallei, is a bacterium commonly found in water, soil and paddy fields in endemic regions [3]. It is highly endemic in northeast Thailand, Malaysia, Singapore and northern Australia [3, 10]. Melioidosis is transmitted via subcutaneous inoculation, inhalation or ingestion of contaminated soil or water with *B. pseudomallei* [4]. In this case, the patient was likely exposed to the pathogen due to her rural agricultural activities.

Clinical diagnosis in this case is challenging because the clinical presentation of sinonasal melioidosis can mimic nasal lymphoma especially NK/T cell lymphoma. They can share similar symptoms such as nasal obstruction, purulent nasal discharge and destruction of the nasal mucosa [7–9]. Similarly, they may be associated with systemic presentation although systemic presentation is less common in nasal lymphoma [7]. Besides, radiological findings for lymphoma include sinonasal mass, sinus opacification and bony erosion which present in this case [9]. With an atypical presentation in this case, we initially suspected that the patient had an underlying immunocompromised status such as diabetes or HIV. Screening tests done were negative. More than 90% of the disease in Malaysia present acutely [4]. The diagnosis of Melioidosis could be easily missed due to non-specific clinical presentation and lack of familiarity with the disease among attending clinicians and laboratory personnel [4]. The gold standard to diagnose Melioidosis is by tissue and bodily fluid culture of *B. pseudomallei* [6]. Fortunately, in our case, tissue culture was positive.

Melioidosis carries a high mortality rate due to early onset of fulminant sepsis [4, 5]. 20% of the cases experienced septic shock with bacteraemia and the mortality rate in septic shock cases are as high as 86–100% [2, 4, 6]. The risk factors for melioidosis includes diabetes, chronic renal disease, tuberculosis, immune disorder and malignancy [2–4].

According to Centres for Disease Control and Prevention (CDC) 2021, treatment for Melioidosis is generally starts with intravenous (IV) Ceftazidime or Meropenem for two to eight weeks depending on extend of infection, followed by oral trimethoprim-sulfamethoxazole or Amoxicillin/clavulanic acid for three to six months [3, 10]. Besides, several

preventive measures are recommended by CDC to those who are at risk of melioidosis, which includes avoid contact with soil and standing water, wearing boots while performing agricultural work. Nonetheless, health care workers can practise standard precautions when treating melioidosis patients.

Summary

Disseminated melioidosis involving sinonasal mimicking nasal lymphoma which carries significant mortality rate. This case highlights the importance of early referral and early diagnosis to avoid delay in appropriate treatment. Also, a low threshold for ICU admission is crucial in preventing mortality. Factors such as young age and no co-morbidity carries positive outcome in the case of melioidosis. Besides, high index of suspicion needed if any atypical presentation of disease.

Declarations

Conflict of interest The authors declare that they have no known competing financial interests or personal relationships that could influence the work of reporting the case.

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