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The Great Mimicker: A Case of Disseminated Melioidosis with Extensive Abscesses and Successful Outcome in an Elderly Male

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Abstract

Introduction: Burkholderia pseudomallei causes melioidosis, a tropical disease common in Southeast Asia [1]. It typically manifests as pneumonia or intra-abdominal abscesses, diagnosed by culture, and treated with specific antibiotics. Objective: To discuss the diagnostic and therapeutic complexities encountered in a case of disseminated melioidosis presenting with atypical presentation of facial swelling and a neck mass in an individual with newly diagnosed type 2 diabetes mellitus. Who has no known exposure to contaminated soil and has not engaged in rice farming. Case presentation: A 62-year-old male presented with facial swelling and an enlarging neck mass. A computed tomography (CT) scan of the neck and chest revealed a rim-enhancing collection in the right lateral cervical region with loculations, extensions, and surrounding inflammatory changes, consistent with abscess formation. Additional rim-enhancing, hypodense collections were observed in the right paratracheal, paraesophageal, and prevertebral regions extending from T1 to T4, indicative of disseminated abscess formation. These imaging findings, in conjunction with systemic manifestations, raised concerns for disseminated infection. Management: The patient underwent serial wound debridement to remove the accessible necrotic tissue and facilitate abscess drainage. Cultures grew B. pseudomallei. Deeper-seated infections were managed conservatively with a four-week course of intravenous meropenem, a first-line therapy for severe melioidosis. This integrated therapeutic strategy, encompassing surgical intervention and prolonged antibiotic administration, is consistent with established clinical guidelines for disseminated melioidosis. Follow-up imaging revealed resolution of the abscesses. Conclusion: This case underscores the considerable diagnostic challenges and intricate clinical course associated with disseminated melioidosis, mandating a heightened index of suspicion, partic-

ularly within endemic geographical areas. Despite the complexity of therapeutic management, achieving a favorable clinical outcome remains a viable possibility.

Keywords

Melioidosis, Burkholderia Pseudomallei, Disseminated Infection, Neck Abscess

1. Introduction

Melioidosis, a significant infectious disease affecting both human and animal populations, exhibits a broad spectrum of clinical manifestations [2]. These range from asymptomatic carriage to localized cutaneous ulcerations or abscesses, chronic pulmonary disease mimicking tuberculosis, and fulminant septicemia characterized by disseminated visceral abscesses. Epidemiological studies in endemic regions, such as northeast Thailand and northern Australia, have demonstrated a strong correlation between melioidosis incidence and periods of high rainfall, with 75% and 81% of cases occurring during the wet season, respectively [3].

The causative bacterium, Burkholderia pseudomallei, is a saprophytic organism prevalent in soil and surface water within endemic areas. Transmission to humans and animals typically occurs through percutaneous inoculation, inhalation of aerosolized bacteria, aspiration of contaminated fluids, or ingestion. Notably, a study conducted in Singapore identified a positive association between melioidosis incidence and increased humidity and rainfall [4]. Furthermore, more recent research from Thailand suggests that ingestion of water contaminated with B. pseudomallei may represent a more frequent route of infection than previously recognized, particularly in endemic regions with untreated water sources [5].

A critical challenge in the management of melioidosis lies in the intrinsic resistance of B. pseudomallei to many commonly prescribed empirical antibiotic regimens used for suspected bacterial sepsis [6]. This inherent resistance underscores the potential for delayed diagnosis and consequently, increased mortality. Therefore, a high index of clinical suspicion is paramount in endemic settings. This case report serves to illustrate the protean and often non-specific clinical presentation of melioidosis and highlights the inherent complexities in the diagnosis and management of disseminated abscess formation associated with this potentially life-threatening infection.

2. Objective

This case report aims to discuss the diagnostic and therapeutic complexities encountered in the management of disseminated melioidosis, specifically as it presented with atypical clinical features of facial swelling and a cervical mass in a 62-year-old male with newly diagnosed type 2 diabetes mellitus, who has no known exposure to contaminated soil and has not engaged in rice farming.

3. Case Presentation

Three weeks prior to admission, the patient noted the insidious onset of a small, erythematous mass in the right lateral cervical region. This initial finding was not associated with fever, purulence, dysphagia, or difficulty in breathing. During the interim, the cervical mass progressively enlarged, reaching a diameter approximating that of a golf ball, accompanied by surrounding erythema. The mass also became fluctuant. The patient subsequently reported dysphagia and odynophagia, while maintaining adequate oral intake. Within the five days preceding hospital admission, the patient experienced a decline in appetite, fever, hoarseness, and worsening dysphagia. The rapid progression of the cervical swelling prompted admission for further evaluation and management.

4. Workups

Workup revealed a significantly elevated white cell count of 31.2×10^9/L. Glycosylated hemoglobin (HbA1c) was elevated at 13.4%, indicating newly diagnosed type 2 diabetes mellitus. The definitive diagnosis of melioidosis was confirmed through the isolation of B. pseudomallei from neck tissue cultures obtained during surgical debridement. Antimicrobial susceptibility testing of the isolate demonstrated susceptibility to meropenem and trimethoprim-sulfamethoxazole, but resistance to ceftazidime and doxycycline

Imaging studies demonstrated extensive abscess formation. As seen in Figure 1, computed tomography (CT) scan of the head and neck revealed a rim-enhancing fluid collection with loculations and surrounding inflammatory changes in the right lateral cervical region, consistent with abscess. The primary collection measured approximately 15.2 × 11.8 × 7.6 cm (craniocaudal × transverse × anteroposterior dimensions) with an estimated volume of 708.8 cm^3. Figure 2 shows a chest CT scan revealed rim-enhancing, hypodense collections with intralesional

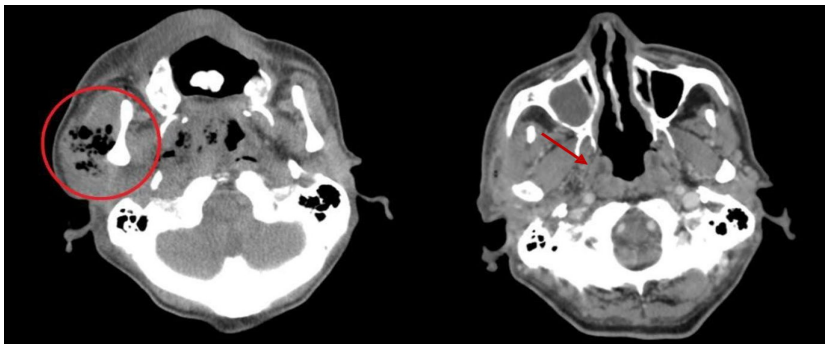

Figure 1. Radiographic comparison of Head and Neck Ct scan: Left panel, image from admission, right panel, image from 4 weeks of antibiotic therapy. On admission noted rim-enhancing collection in the right lateral cervical region with loculations and surrounding inflammatory changes, consistent with abscess formation. The primary collection measured approximately 15.2 × 11.8 × 7.6 cm (craniocaudal × width × anteroposterior), with an estimated volume of 708.8 cc. After 4 weeks of Intravenous antibiotics, complete resolution of rim-enhancing collection in the right lateral cervical region involving the right submandibular, masticator and parotid spaces.

air foci located in the right paratracheal and paraesophageal regions at the T1 to T4 vertebral levels. This collection measured 5.3 × 1.7 × 2.7 cm (craniocaudal × transverse × anteroposterior dimensions), with a calculated volume of 12.7 cm^3. Additional hypodense collections in the posterior paraesophageal/prevertebral region at the T1 to T4 vertebral levels measured 7.0 × 3.2 × 0.9 cm, with an approximate volume of 10.5 cm^3 as shown in Figure 3.

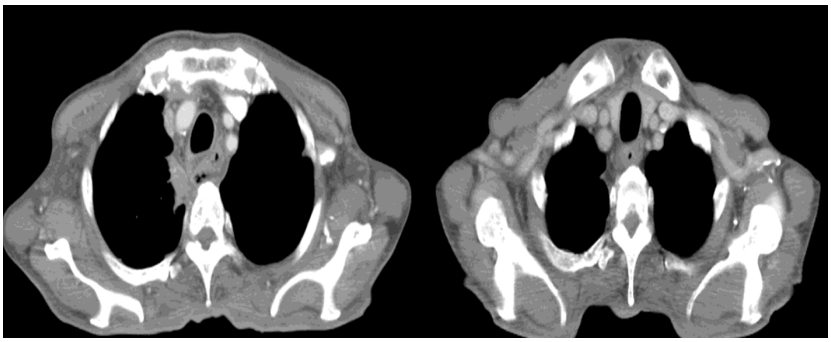

Figure 2. Radiographic comparison of Chest CT scan: Left panel, image from admission, right panel, image from 4 weeks of antibiotic therapy. On Admission, rim-enhancing, hypodense collections with intralesional air foci located in the right paratracheal and paraesophageal regions at the T1 to T4 levels. The collection measured 5.3 × 1.7 × 2.7 cm (craniocaudal × width × anteroposterior), with a calculated volume of 12.7 cc. After 4 weeks of Intravenous antibiotics,with complete resolution of rim-enhancing collection in the right paratracheal and paraoesophageal regions.

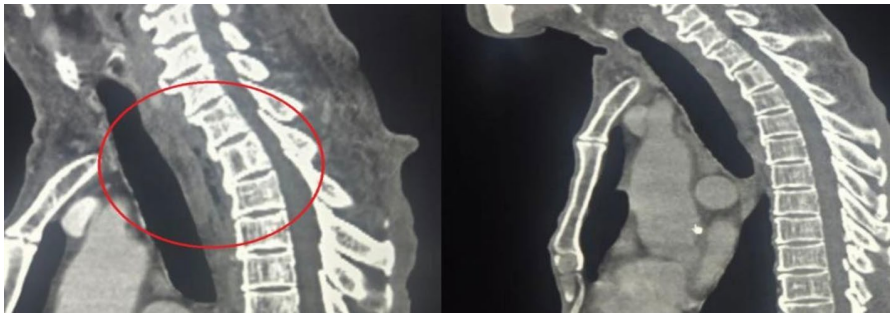

Figure 3. Radiographic comparison of Chest CT scan: Left panel , image from admission, right panel, image from 4 weeks of antibiotic therapy. On admission, Hypodense collections in the posterior paraesophageal/prevertebral region at the T1 to T4 levels. Complete resolution of hypodense collection in the posterior paraesophageal/prevertebral region after 4 weeks of IV antibiotics.

5. Management

The patient was given intravenous meropenem at a dosage of 1 gram every 8 hours for a duration of four weeks. As seen in Figure 4, surgical management involved serial wound debridement of the right lateral cervical abscess. The deeper-seated collections in the paratracheal, paraesophageal, and prevertebral regions were managed conservatively with antibiotic therapy, as the patient declined an open thoracic surgical procedure for drainage. Glycemic management was initiated promptly upon detection of a markedly elevated HbA1c of 13.4%. In coordination

with the Endocrinology service, intensive glucose control was commenced using long-acting insulin glargine. Capillary blood glucose levels were monitored every eight hours, with insulin dose adjustments based on pre-meal values and the patient's overall clinical condition. Tight glycemic control was maintained to enhance immune function and promote optimal wound healing.

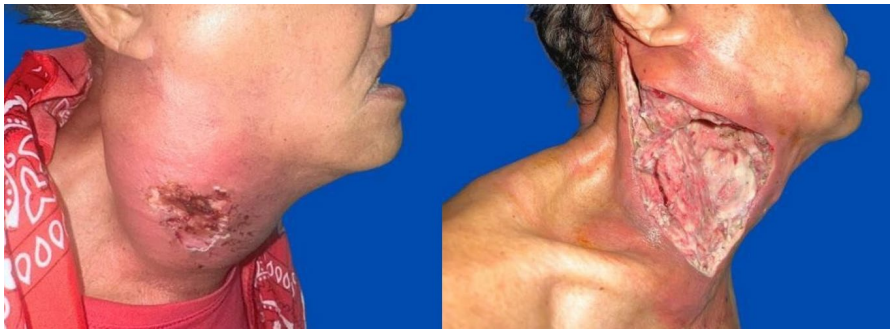

Figure 4. Gross comparison of Cervical Mass: Left panel, image from admission, right panel, image from post surgical debridement. On admission, a large ulcerated and necrotic lesion on the right lateral neck with surrounding erythema and swelling, post surgical debridement, with extensive tissue loss after removal of necrotic tissues.

Following seven days of intravenous antibiotic administration, resolution of fever and leukocytosis was observed. A follow-up CT scan performed after four weeks of intravenous antibiotic therapy demonstrated complete resolution of the previously identified abscesses. As seen in Figure 5, the surgical site exhibited favorable granulation tissue formation, and the patient was subsequently discharged from the hospital. Eradication therapy with oral trimethoprim-sulfamethoxazole at a dosage of 320 mg/800mg (two tablets every 12 hours) was administered for a total duration of 12 weeks.

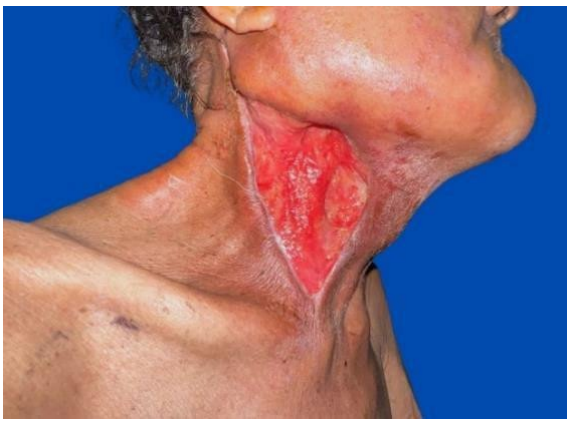

Figure 5. Gross feature of cervical area after four weeks of intravenous antibiotics and serial surgical debridement good granulation and absence of necrotic tissues and signs of inflammation.

6. Discussion

Disseminated melioidosis, an infection caused by the Gram-negative bacterium B.

pseudomallei, presents significant management challenges owing to its diverse clinical manifestations and intrinsic resistance to numerous commonly employed antibiotics [7] [8]. The case at hand, characterized by a progressively enlarging cervical mass and systemic symptoms, effectively demonstrates the unpredictable nature of this disease, frequently termed "The Great Mimicker" due to its capacity to simulate a wide array of infectious and non-infectious entities [9].

Disseminated melioidosis represents a severe form of the infection characterized by the presence of abscesses in multiple organs or sites beyond the primary focus of infection (e.g., lungs, skin). This pattern of spread underscores the hematogenous or lymphatic dissemination of B. pseudomallei from an initial site of entry, which is often cutaneous inoculation or inhalation. The clinical manifestations of disseminated melioidosis are highly variable and can involve virtually any organ system, leading to a complex array of signs and symptoms. Common sites of metastatic abscess formation include the liver, spleen, prostate gland, and musculoskeletal system, but involvement of the head and neck region, as seen in the present case, is also documented. Head and neck infections, though rare, are documented [10], while intraosseous abscesses have also been reported [11].

The pathogenesis of disseminated melioidosis involves the ability of B. pseudomallei to survive and multiply intracellularly within macrophages and neutrophils, facilitating its spread throughout the body. Host factors, particularly the presence of underlying conditions such as diabetes mellitus, chronic kidney disease, and immunosuppression, significantly increase the risk of developing disseminated disease and are associated with poorer clinical outcomes [12]. The high mortality rate associated with disseminated melioidosis necessitates prompt diagnosis and aggressive treatment with appropriate antimicrobial agents and, often, surgical drainage of abscesses [13]. The prolonged duration of treatment required to eradicate the infection and prevent relapse further highlights the challenges in managing this severe form of melioidosis [9].

Within the Philippines, the earliest documented instance of melioidosis dates back to 1948, involving a patient who manifested with weight loss and pneumonia [14]. Subsequent studies have further elucidated the clinical landscape of melioidosis in the country. For instance, a case series by Velez [15] described the varied presentations of melioidosis in Filipino patients, highlighting pulmonary involvement, skin and soft tissue infections, and visceral abscesses as common manifestations. More recently, Maningas *et al.* (2018) [16] reported a cluster of melioidosis cases following a flooding event, emphasizing the environmental link to infection and the potential for outbreaks. Furthermore, the case by Perez *et al.* (2021) [17], detailing disseminated melioidosis with splenic, musculoskeletal, and high anal fistula involvement, underscores the continued occurrence of this complex disease with atypical presentations in the Philippines. These local studies, alongside global literature, emphasize the need for a heightened index of suspicion for melioidosis, particularly in endemic regions like the Philippines, to facilitate timely diagnosis and appropriate management.

This case further emphasizes the unpredictable clinical trajectory of disseminated melioidosis, wherein the patient presented with a progressively enlarging cervical mass accompanied by systemic manifestations. The propensity for chronic or latent infections, followed by systemic dissemination and subsequent abscess formation in diverse anatomical locations such as the head, neck, and even osseous structures [13]. Such multifaceted presentations necessitate comprehensive diagnostic evaluations, including advanced imaging modalities, to delineate the extent of the infection. Notably, diabetes mellitus is an established and significant risk factor for the acquisition and severity of melioidosis, as hyperglycemia is known to impair the host's innate immune response, thereby increasing susceptibility to B. pseudomallei infection. The well-established association between diabetes mellitus and an increased susceptibility to melioidosis aligns with global epidemiological data, as reviewed by Chakravorty [13] underscoring the importance of early screening for underlying metabolic conditions in individuals residing in or traveling to endemic regions. In the case presented, the patient's previously undiagnosed type 2 diabetes likely played a substantial role in the development and dissemination of the melioidosis.

Melioidosis is an endemic but likely underreported disease in the Philippines However, in this particular case, the patient has no known exposure to contaminated soil and has not engaged in rice farming. While exposure to soil and water, particularly in agricultural settings like rice fields, is a significant risk factor highlighted in a case-control study during a 2019 outbreak in Isabela Province [18] and consistently identified in other endemic regions like Thailand the absence of this typical exposure in our patient suggests other potential, less common routes of infection or the possibility of an unidentified environmental source. Potential exposures include contact with contaminated floodwaters, soil handling at construction sites, unprotected gardening activities, and immersion in untreated natural water bodies. Despite the usual association with agricultural work seen in local case reports and outbreaks [19] [20]. The occurrence of melioidosis in individuals without such direct exposure underscores the need to consider alternative transmission pathways, especially when coupled with predisposing factors like diabetes mellitus, the most common comorbidity observed in Filipino melioidosis patients.

The timely and accurate diagnosis of disseminated melioidosis necessitates a multimodal approach integrating both laboratory and advanced imaging techniques. Isolation and identification of B. pseudomallei from clinical specimens, such as tissue cultures obtained during surgical debridement as demonstrated in this case, remains the definitive diagnostic standard. Complementary to microbiological confirmation, advanced radiological imaging, including CT and magnetic resonance imaging, plays a crucial role in delineating the extent and location of deep-seated abscesses, as highlighted by Soo *et al.* (2022) [12] in their discussion of imaging's utility in guiding both medical and surgical management strategies for melioidosis. Effective therapeutic management of disseminated melioidosis

frequently requires a synergistic combination of prolonged intravenous antibiotic therapy and surgical intervention. Loh *et al.* (2017) [10] specifically addressed severe manifestations of melioidosis in the head and neck region, including extensive facial soft tissue infections and septic metastatic foci, emphasizing the critical role of combined surgical and prolonged antibiotic approaches in achieving favorable outcomes. Intravenous meropenem, as administered in this case, is recognized as a first-line antibiotic for the treatment of severe melioidosis due to its broad-spectrum activity and efficacy against B. pseudomallei [1]. Following the initial intensive intravenous phase, a prolonged eradication phase with oral antibiotics, such as trimethoprim-sulfamethoxazole (cotrimoxazole), is essential to minimize the risk of relapse [8]. In cases involving substantial tissue destruction and abscess formation, surgical drainage and debridement are critical adjunctive measures to reduce the bacterial burden and facilitate source control, as further supported by the findings of Loh *et al.* (2017) [10].

7. Conclusion

This case emphasizes the necessity for increased diagnostic vigilance regarding melioidosis, especially in endemic regions and among individuals with predisposing factors such as diabetes mellitus. Melioidosis, a multifaceted disease with protean clinical manifestations due to its potential to involve multiple organ systems, is susceptible to misdiagnosis and delayed treatment initiation. Prompt and precise identification via culture and imaging modalities is crucial for effective clinical management. Optimal outcomes necessitate a combination of targeted antimicrobial therapy and surgical intervention, particularly for substantial or deep-seated abscesses. Timely source control through surgical drainage, coupled with appropriate and prolonged antibiotic administration, has demonstrated significant improvement in recovery rates. Furthermore, this case underscores the importance of enhanced awareness among healthcare professionals to mitigate diagnostic delays and ensure optimal management strategies for melioidosis. Continued research is imperative to refine treatment protocols and develop improved approaches for managing disseminated abscesses associated with this challenging infection.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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