

## Case Report

### **Melioidosis presenting with periorbital cellulitis and rhinosinusitis.**

Udayani LADN<sup>1</sup>, Dumingoarachchi D<sup>2</sup>

<sup>1</sup> Senior Registrar in ENT, Lady Ridgeway Hospital (LRH), Sri Lanka

<sup>2</sup> Consultant ENT and Head & Neck Surgeon, Teaching Hospital Anuradhapura (THA), Sri Lanka

#### **Abstract**

Melioidosis is an infectious disease caused by the bacteria *Burkholderia pseudomallei*. It has a variety of clinical manifestations ranging from localized abscess, to severe pneumonia to fatal septicaemia. Sinonasal and orbital manifestations of Melioidosis are rare <sup>[1]</sup>. This is the first reported case of Melioidosis presenting with orbital cellulitis and rhinosinusitis in Sri Lanka. A 42-year-old patient with diabetes mellitus presented with pyrexia of unknown origin, multiple abscesses in the leg, periorbital cellulitis and rhinosinusitis. Initial investigations did not reveal a diagnosis. Eventually it was diagnosed as Melioidosis with a positive blood culture and successfully managed with long term antibiotics.

#### **Conclusion**

Early identification and aggressive management is needed to reduce the morbidity and mortality of Melioidosis. Awareness amongst clinicians of different specialties is important as this disease has variety of manifestations.

**Keywords:** Melioidosis, Rhinosinusitis and orbital complications

**Copyright:** © 2021 Udayani LADN et al

This is an open access article distributed under the [Creative Commons Attribution License \(CC BY 4.0\)](https://creativecommons.org/licenses/by/4.0/)



This license lets others distribute, remix, tweak, and build upon the work, even commercially, as long as they credit the original author for the creation.

**Funding:** None

**Competing interest:** None

**Correspondence:** Udayani LADN ([nadeejalad@gmail.com](mailto:nadeejalad@gmail.com))

Accepted Date: 22<sup>nd</sup> Feb 2021

Published Date: 12<sup>th</sup> March 2021

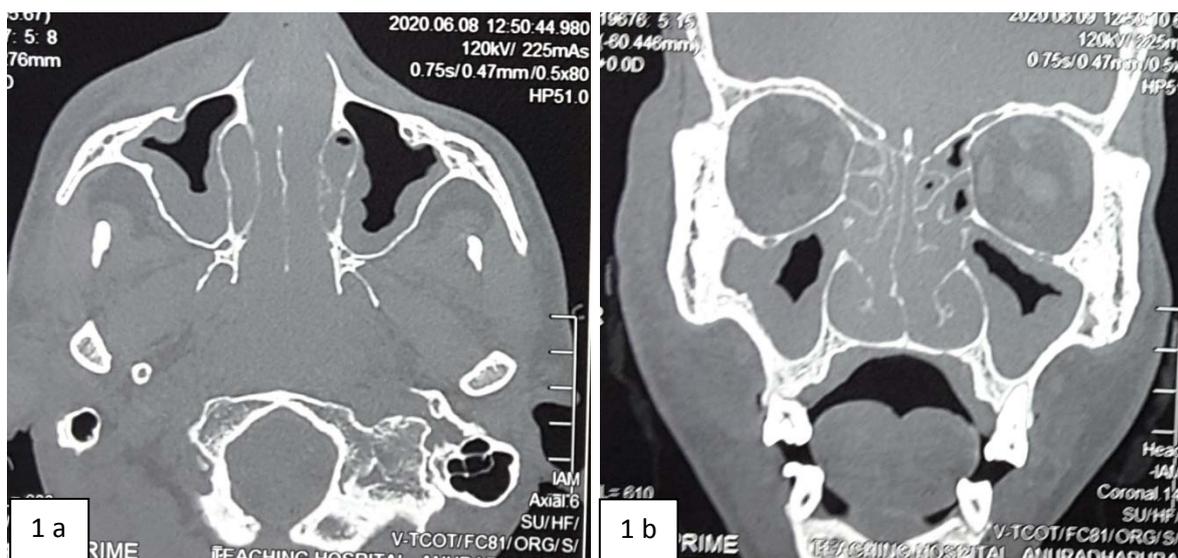
## Introduction

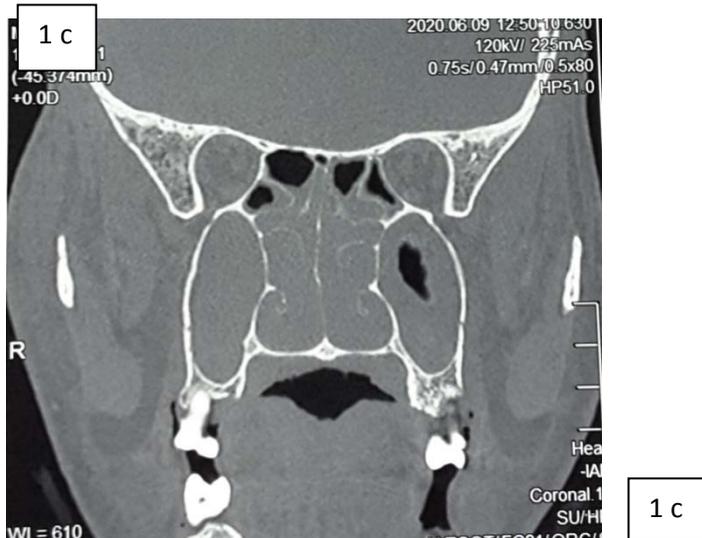
The causative organism of Melioidosis is *Burkholderia pseudomallei* [2], which is a Gram-negative soil- and water associated pathogen. [3] Humans are infected by traumatic inoculation of bacteria from soil or rarely by inhalation or ingestion [4,5]. The disease is endemic to southeast Asia and northern Australia [6,7] although sporadic cases are reported from Sri Lanka. The disease is more common amongst patients with diabetes mellitus, renal disease, and alcoholism [8]. Melioidosis is an infectious disease with a variety of clinical manifestations [1]. Manifestations may range from localized abscess, to severe pneumonia to fatal septicaemia [9]. Only a few cases have been reported with rhinosinusitis and orbital complications.

## Case report

A 42-year-old male from Galgamuwa, with a past history of diabetes mellitus presented to a medical unit with on and off fever for two months' duration. He had two recent hospital admissions with fever and managed as urinary tract infections with intravenous and oral antibiotics. In the current admission he had high fever. Whilst in the ward he developed abscesses in the leg and chest wall cellulitis which were managed with antibiotics and drainage of leg abscesses. While he was being investigated for pyrexia of unknown origin, he developed a nasal congestion, rhinorrhoea, headache and right sided facial and periorbital swelling. His vision remained normal with mild pain on eye movements. He was then referred to the ENT team.

On examination he was febrile. Anterior rhinoscopy showed a congested nasal cavity. There was right sided periorbital cellulitis. His vision and eye movements were normal. Rigid nasal endoscopic examination revealed congested and inflamed nasal mucosa in his nasal cavity with a necrotic patch in the posterior nasal septal mucosa. Full blood count showed a neutrophil leucocytosis, ESR was 102 mm/1<sup>st</sup> hour, CRP was 206 g/dl and renal functions were normal. As he was a diabetes mellitus patient with fever of unknown origin and sinonasal symptoms with eye swelling, our main differential diagnosis was invasive fungal sinusitis. We requested an urgent Contrast enhanced (CE) CT of the nose and paranasal sinuses and it revealed soft tissue density material in bilateral frontal, ethmoid, sphenoidal and maxillary sinuses. Extensive soft tissue material noted occluding bilateral nasal passages. Soft tissue stranding's were extending outside the maxillary sinus and this raised the suspicion for invasive fungal sinusitis.





Picture 1  
1a (Axial  
1b (Coron  
1c (Coron

He then underwent an urgent endoscopic sinus surgery, bilateral middle meatal antrostomy, ethmoidectomy and sphenoidotomy. The mucosa of sinus cavities was normal. There was a necrotic mucosa in posterior nasal septum. The necrotic mucosa was removed and it was sent for urgent fungal studies and culture, histology and bacterial culture. There were no fungal filaments in the direct smear of this tissues. There were no bacteria or fungal growth in the cultures. Histology only revealed acute inflammatory changes and did not demonstrate any fungal filaments. Invasive fungal sinusitis was excluded.

Patient was started on intra venous Meropenem, nasal douching and Betamethasone nasal drops. His periorbital cellulitis responded to the intra venous antibiotics. There was a growth in blood culture and it was confirmed as *Burkholderia* by the Medical research institute (MRI) Borella. *Burkholderia* antibody was done and it was positive. Intravenous Meropenem was continued for 60 days. He fully recovered and was discharged on oral Co-Trimaxazole 480mg twice day and we planned to continue this for six months.

## Discussion

Melioidosis is an infectious disease caused by gram negative bacilli *Burkholderia pseudomallei*. It is an endemic disease in Southeast Asia; Australia; Central, West, and East Africa; India; the Middle East; and China and sporadic cases reported worldwide. In humans, infection is mainly occurs by contamination of skin abrasions or burns, ingestion, or inhalation but not directly from infected animals or other humans. The disease is more common amongst patients with diabetes mellitus, chronic renal disease, immune suppression, chronic lung disease, and malignancies.

Melioidosis has variety of manifestations. Presentation may be acute or chronic. Acute pulmonary infection is the most common form of presentation. Suppurative infections can occur in almost any organ, but most common sites are inoculation sites in the skin, associated lymph nodes, or lung. Acute septicaemic Melioidosis is usually fatal. Sinonasal and orbital presentations are rarely documented in the literature and there have been no reported cases in Sri Lanka.

Melioidosis is diagnosed by isolating *Burkholderia pseudomallei* from blood, urine, sputum, throat swabs, skin lesions, or abscesses; or by detecting an antibody response to the bacteria. Melioidosis can be successfully treated most of the time with the appropriate antibiotics. Treatment is generally starts with intravenous antibiotics followed by a 3-6 month course of oral antibiotics. Intravenous ceftazidime, meropenem, imipenem, piperacillin are effective in the treating this condition<sup>[10]</sup>. Oral therapy can be continued with Trimethoprim/sulfamethoxazole or Doxycycline.

## **Conclusion**

Melioidosis is a disease with significant morbidity and mortality. Early identification and aggressive management is needed to reduce the morbidity and mortality. Awareness among clinicians of different specialties is important as this disease has variety of manifestations. Melioidosis should be considered in the differential diagnosis of rhinosinusitis in an immunocompromised patient after excluding other diseases like invasive fungal sinusitis.

## References

1. Wong P.K, Ng P.H.; Melioidosis presenting with orbital cellulitis; Singapore med j 1996; Vol 37; 220-221
2. Perera GND, Dias LD, Kulatunga A, Corea E, Masakorala J, A case report of Melioidosis. Srilankan Journal of Infectious Diseases, 2012;2(1):47-51  
DOI: <http://doi.org/10.4038/sljid.v2i1.3795>
3. Currie B.J. Melioidosis an important cause of pneumonia in residents of and travelers returned from endemic regions; European respiratory journal Sep 2003, 22(3) 542-550,  
DOI: <https://doi.org/10.1183/09031936.03.00006203>
4. Corea E ; Thevanesam V; Perera S; Jayasinghe I, Ekanayake A, Masakorala J ; Melioidosis in Sri Lanka: an emerging infection ;*Sri Lanka Journal of Infectious Diseases* 2012 Vol.1(2):2-8,  
DOI: <http://dx.doi.org/10.4038/sljid.v2i1.3801>
5. Rebecca SinMei Lim, Sam Flatman, and Markus C. Dahm; Sinonasal Melioidosis in a Returned Traveller Presenting with Nasal Cellulitis and Sinusitis; Hindawi Publishing Corporation; Case Reports in Otolaryngology; Volume 2013, Article ID 920352,  
<https://doi.org/10.1155/2013/920352>
6. Allen C. Cheng, Bart J. Currie; Melioidosis: Epidemiology, Pathophysiology, and Management ;*Clinical microbiology reviews*, Apr. 2005, p. 383–416, doi: [10.1128/CMR.18.2.383-416.2005](https://doi.org/10.1128/CMR.18.2.383-416.2005)
7. Huang, Wy., Wu, G., Chen, F. *et al.* Multi-systemic melioidosis: a clinical, neurological, and radiological case study from Hainan Province, China. *BMC Infect Dis* 18, 649 (2018),  
<https://doi.org/10.1186/s12879-018-3569-8>
8. Currie BJ, Fisher DA, Howard DM, et al. Endemic melioidosis in tropical northern Australia: a 10-year prospective study and review of the literature. *Clin Infect Dis*. 2000;31(4):981–6, doi: 10.1086/318116.
9. Leelarasamee A., Bovornkitti S., Melioidosis: Review and Update, *Reviews of Infectious Diseases*, Volume 11, Issue 3, May 1989, Pages 413–425, <https://doi.org/10.1093/clinids/11.3.413>
10. Inglis TJJ; Challenges in diagnosis and treatment of melioidosis ;*Sri Lanka Journal of Infectious Diseases* 2012 Vol.1(2); 52-53, DOI: <http://dx.doi.org/10.4038/sljid.v2i1.4067>