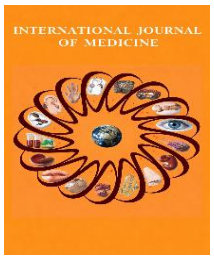
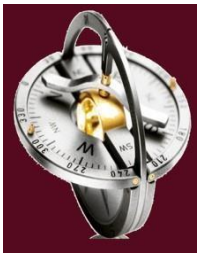


International Journal of Medicine
ISSN (P): 1468-3814; ISSN (e): 2667-7008

https://www.theinternationalmedicine.com



Volume: 5(1) 2023

Case Report

Unusual cutaneous miliary tuberculosis manifesting as periorbital necrotizing fasciitis: Pitfalls and pearls

Mohamad Aizat Rosli[1,2,3], Shawaltul Akhma Harun nor Rashid[3], Ahmad Sukari Halim[1,2]

[1]*Hospital Universiti Sains Malaysia, Medical Campus. Universiti Sains Malaysia, Kota Bharu, Kelantan, Malaysia*

[2]*Reconstructive Sciences Unit, School of Medical Sciences, Medical Campus. Universiti Sains Malaysia, Kota Bharu, Kelantan, Malaysia*

[3]*Plastic and Reconstructive Unit, Hospital Raja Perempuan Zainab II, Kota Bharu, Kelantan, Malaysia*

Received: 11 January 2023 / Accepted: 11 February 2023

Abstract

Periorbital Necrotizing fasciitis (NF) is a severe infection marked by progressive and rapid necrosis of superficial fascia, often caused by Group A *Streptococcus*. However, there are increasing reports of rare involvement of *Mycobacterium tuberculosis* in its pathogenesis. Failure to identify and commence appropriate treatment based on its pathogenesis can result in extensive facial disfigurement, blindness, meningitis, brain abscess, septicemia, and even death. This case report aims to highlight a number of salient points in managing a rare case of cutaneous miliary tuberculosis presenting as periorbital necrotizing fasciitis.

Keywords: bacterial skin diseases; miliary tuberculosis; necrotizing fasciitis

1. Introduction

Necrotizing fasciitis (NF) is a severe infection marked by progressive and rapid necrosis of superficial fascia, along with the involvement of the overlying skin, subcutaneous fat, and underlying muscles [1]. It is rare in the head and neck regions, particularly in the preorbital area, due to the face's good vascularity [2,3]. Group A *Streptococcus* is the most familiar causative agent in facial NF, often precipitated by trauma, odontogenic infection, or surgery [4,5]. However, there are also reports, albeit scarce, describing the rare involvement of *Mycobacterium tuberculosis* in its pathogenesis.

Early detection is a challenge as many clinicians are not familiar with NF. In its early stage, NF is often mistaken for cellulitis. The critical feature of NF is rapid tissue necrosis [6,7]. The right diagnosis should be made so that treatment can be administered on time to prevent further tissue damage [5]. A high index of suspicion is required to prompt rapid treatment. More importantly, the right etiologies and organisms must be correctly identified and addressed, especially in a complicated case where different subgroups of organisms are involved.

Failure to commence appropriate treatment can result in extensive facial disfigurement, blindness, meningitis, brain abscess, septicemia,

Address for Correspondence: Ahmad Sukari Halim, Hospital Universiti Sains Malaysia, Jalan Raja Perempuan Zainab II, Kubang Kerian, 16150 Kota Bharu, Kelantan, Malaysia. E-mail: ashalim@usm.my

DOI: 10.5455/im.34116

and even death [8]. Furthermore, the insidious infection caused by microorganisms such as *Mycobacterium tuberculosis* needs to be detected so that antituberculotic drugs can be administered.

This case report will highlight several salient points in managing a rare case of cutaneous miliary tuberculosis presenting as periorbital necrotizing fasciitis presented to Hospital Raja Perempuan Zainab II, Kelantan, Malaysia.

2. Case Report

A 33-year-old Malay man with no known medical illness presented to our casualty with a one-day history of bilateral eye swelling following a bite from an unknown insect in December 2019. He noticed a wheal-like lesion measuring 0.5 cm over his right medial canthal region, which rapidly progressed to a tender diffuse swelling over the right periorbital region [Figure 1, left picture]. No fever, blurry of vision, headache, or any constitutional symptom was reported. The ophthalmology team's initial examination only revealed swelling and erythema of bilateral upper and lower eyelids, with bilateral chemosis and injected conjunctiva. The vision was otherwise remarkable. His blood investigations showed a high total white cell count; 17 x10^9 /L with a neutrophil predominance (91%). C-Reactive protein was 283 mg/L. He was treated as bilateral preorbital cellulitis and commenced on cloxacillin 1 g 6-hourly. Upon further history, he had a history of weight loss of about 40 kg over six months, and he was already worked out and cleared from tuberculosis (TB) at another health center. Repeated TB workout, viral and autoimmune screenings were taken to rule out immunodeficiency state.

Despite antibiotics, the swelling worsened and spread to the neck region. A high spike of fever was also recorded in the ward (38.5 degrees Celsius). Multiple necrotic patches progressively developed in his right upper and lower eyelids region [Figure 1, middle picture]. Surrounding skin was also seen to be more erythematous and indurated, with pus discharge. The antibiotic was escalated to IV ceftriaxone 2 g daily and IV metronidazole 500 mg 8-hourly. A contrast-enhanced computed tomography (CT) scan of the brain, orbit, head, and neck [Figure 1, right picture] showed bilateral peri-septal soft thickening with fat streakiness, mostly on the right side. There was also increased thickness of subcutaneous fascia and fat with fat stranding from right cheek till neck, with multiple enlarged nodes seen suggesting extensive subcutaneous tissue inflammation. Brain and bilateral globes were normal. Multiple lung nodules were seen bilaterally, which was likely infective in origin, hence pulmonary military TB was also considered. Swab for culture and sensitivity was taken from the wound grew Hemolytic Group *Streptococcus pyogenes*, while the blood culture taken initially did not grow any bacteria. The viral and autoimmune screening were negative. The sputum smear for Acid Fast Bacilli (AFB) was also negative. The patient was then treated for necrotizing fasciitis of the bilateral preorbital region.

Figure 1. The appearance of the necrotizing fasciitis upon initial presentation, as seen before admission (left), during admission (middle), and in the CT scan (right).

Wound debridement was performed twice due to inadequate response to antibiotic treatment. Necrotic patches were still confined within the original areas after demarcation [Figure 2, left picture]. However, a considerable amount of pus and tissue necrosis were seen beyond the original area, with an insinuation of pus between the subcutaneous layer and muscles along the superficial muscular aponeurotic system's fascia. The right orbicularis oculi, procerus, levator palpebrae superioris muscles appeared unhealthy and necrosed, however tarsal plate and septum were still intact [Figure 2, middle and right pictures]. Debridement was done conservatively to preserve critical

anatomical landmarks and avoid excessive disfigurement while ensuring an adequate amount of necrotic tissue and pus removed. Tissue culture taken intraoperatively grew *Candida albicans*, hence fluconazole was added to his treatment regime. An MRI of the brain, the orbit was done post-operatively as the patient had progressive bilateral blurry vision. The MRI showed a thin enhancement along the right optic nerve suggestive of optic neuritis, however bilateral eye globes and brain were normal.

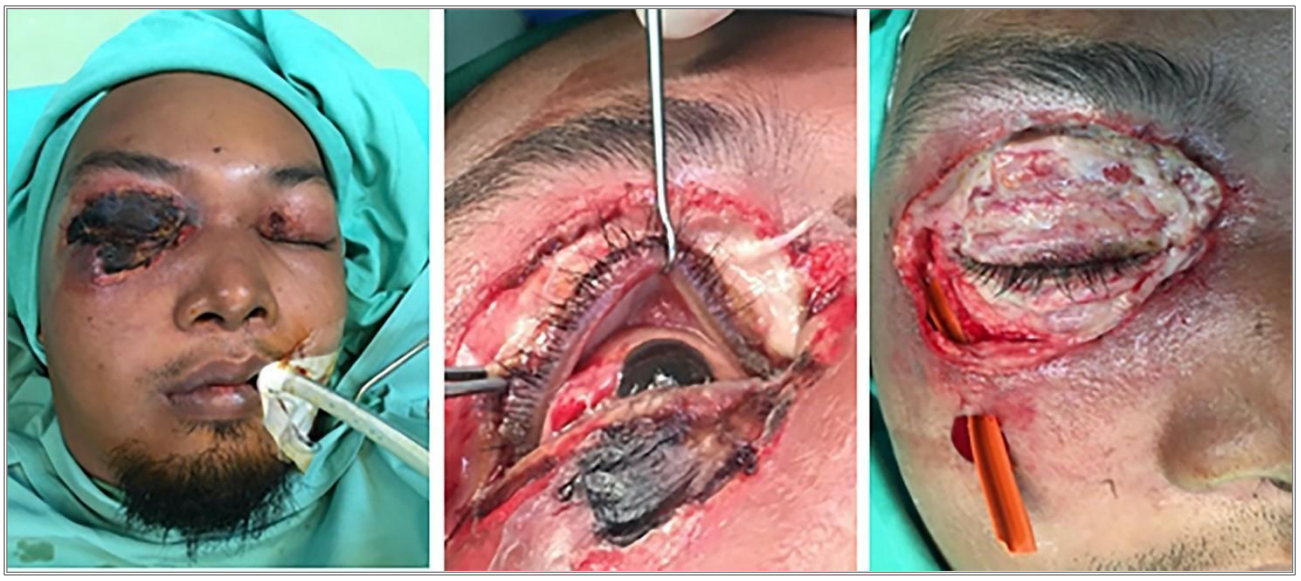


Figure 2. The appearance before surgery: the necrotic area was more demarcated (left). Intraoperatively, the right orbicularis oculi, procerus, levator palpebrae superioris muscles appeared unhealthy, sparing the tarsal plate and septum (middle, left).

He subsequently lost his bilateral vision due to optic neuritis. After series of daily wound dressing and flushing, the defect was then reconstructed with a skin graft two weeks later as he did not agree to any major soft tissue reconstruction [Figure 3, left picture]. A few tissue samples were sent for histopathology examination (HPE), tissue Acid Fast Bacilli (AFB), and tissue *Mycobacterium tuberculosis* (MTB) culture intraoperatively. The HPE result revealed a suppurative granulomatous inflammation with epithelioid histiocytes, central suppuration, and multinucleated giant cells. The tissue AFB taken was negative for tuberculosis, however, the tissue MTB culture was positive for *Mycobacterium tuberculosis*. Antituberculotic drugs consisting of rifampicin, pyrazinamide, ethambutol, and isoniazid were then commenced immediately. Due to ongoing cutaneous tuberculosis infection, only 50% of the graft was viable [Figure 3, middle picture]. An evisceration of the right eye globe had to be undertaken due to the infection, exposing the raw conjunctiva, as seen in the latest picture [Figure 3, right picture]. Otherwise, the periorbital wound improved with regular dressing. Unfortunately, despite evading more serious sequela such as septicemia, meningitis, and death, this condition still led him to bilateral eye blindness.

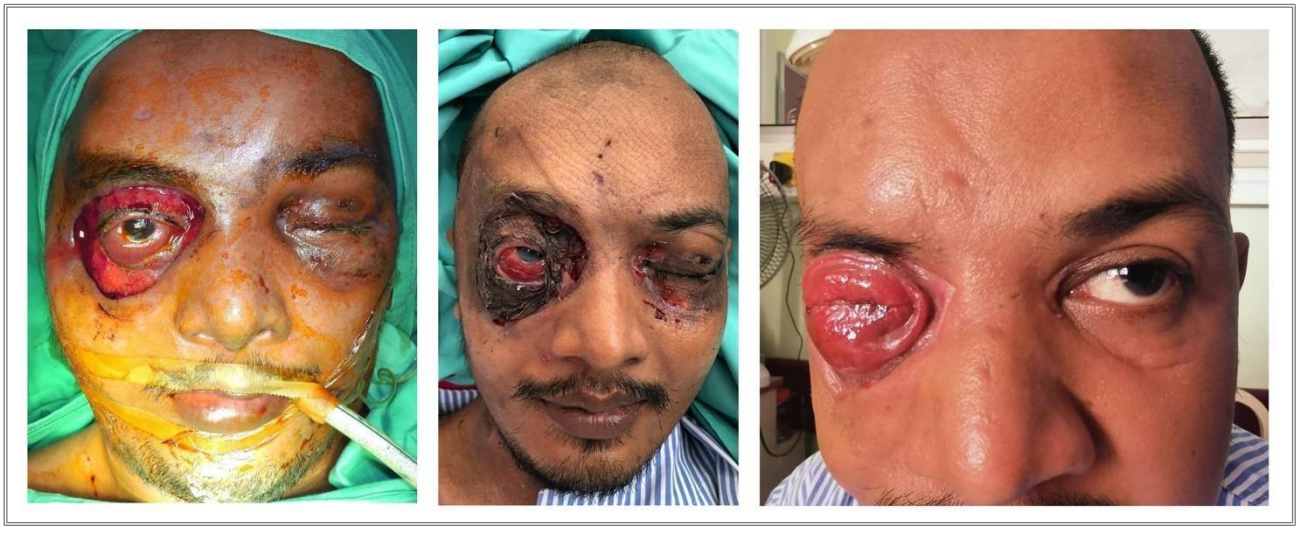


Figure 3. The wound before and after skin graft; however, it was complicated with graft loss due to unresolved infection (left, middle). The appearance post evisceration (right).

3. Discussion

This report highlights an atypical presentation of periorbital NF caused by cutaneous military tuberculosis. In general, NF can be divided into two types. Type 1 is caused by non-group A, polymicrobial infection from obligate anaerobic and aerobic bacteria. Its prevalence is slightly lower than type 2, commonly seen in patients with comorbidities after surgical procedures [4]. The microorganisms involved in type 1 include *Staphylococcus aureus*, *Streptococcus spp*, *Escherichia Coli*, *Enterococcus, Clostridium spp*, and few others. Some have even reported fungi infection in immunocompromised patients. Type 2 is the more prevalent; it is a monomicrobial infection often caused by group A *Streptococcus* in healthy patients with any comorbidity [4]. However, few variations exist, including rare infections caused by *Mycobacterium tuberculosis*.

As portrayed in this case report, tuberculosis was already considered upon the initial presentation. However, due to a negative sputum smear for AFB and a lack of respiratory symptoms, tuberculosis was briefly dismissed. An intravenous broad-spectrum empirical antibiotic was administered and was later converted to a more specific antibiotic based on his tissue culture sensitivity. His first two wound cultures grew Hemolytic Group A *Streptococcus pyogenes* and *Candida albicans*, respectively, the commonly known causes of NF. No specific tissue specimen was sent for HPE, AFB, or MTB culture at that time, as the etiology was already quite clear to us.

However, we noticed that he only had a partial response to the initial antibiotics and surgical debridement. The positive MTB culture result and appearance of granulomatous inflammation in the HPE result sent in the subsequent debridement made us realized that we had missed the correct diagnosis by an oversight. It was assumed that this NF's cause was solely from the Hemolytic Group A *Streptococcus pyrogens*, where in reality, it might have been caused by multiple concomitant etiologies. Based on the evidence, we could conclude that this particular NF's etiology was, in fact, due to *Mycobacterium tuberculosis*. The *Streptococcus pyogenes* and *Candida albicans* isolated initially were presumably opportunistic secondary infections of the local wound.

The lack of awareness that *Mycobacterium tuberculosis* can cause necrotizing fasciitis has misguided us during the initial management, which has resulted in a delay in commencing antituberculotic drugs. It is also worth noting that the lung nodules seen in the earlier CT scan should have alerted us to the possibility of dealing with sputum smear-negative tuberculosis, however, we were blinded by the positive culture results the two microorganisms described previously. This circumstance clearly shows that awareness is vital to facilitate earlier detection and commencement of antituberculotic drugs.

The importance of early debridement must be addressed as well. Based on a study by Lazzeri et al., urgent debridement is crucial to reduce morbidity and mortality. Debridement of visible necrotic tissue is necessary to remove the foci of infection and bacterial load [8]. As NF tracks along avascular fascial tissue planes, intravenous antimicrobial alone is inadequate due to restricted access. Early surgical intervention is also crucial to minimize further skin necrosis by preventing thrombosis of cutaneous perforators [9].

3.1. Conclusion

Necrotizing fasciitis of the periorbital region is rare and life-threatening. Rapid diagnosis and intervention should be made to prevent local tissue damage and systemic organ dysfunction. As it may be caused by an uncommon organism such as *Mycobacterium tuberculosis*, early detection can only be facilitated by having awareness and a high suspicion index.

Conflict of interest

The authors declare no conflict of interest.

Funding

This paper did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sector.

References

1. Gurdal C, Balkan H, Sarac O, Seven E, Yenidunya MO, Kutluhan A, et al. Periorbital necrotizing fasciitis caused by community-associated methicillin-resistant Staphylococcus aureus periorbital necrotizing fasciitis. Orbit 2010;29:348-50.
2. Bisno AL, Stevens DL. Streptococcal infections of the skin and soft tissues. N Engl J Med 1996;334:240-5.
3. Lazzeri D, Lazzeri S, Figus M, Tascini C, Bocci G, Colizzi L, et al. Periorbital necrotising fasciitis. Br J Ophthalmol 2010;94:1577-85.
4. O'Loughlin RE, Roberson A, Cieslak PR, Lynfield R, Gershman K, Craig A, et al. The epidemiology of invasive group A streptococcal infection and potential vaccine implications: the United States, 2000-2004. Clin Infect Dis 2007;45:853-62.
5. Wolf H, Rusan M, Lambertsen K, Ovesen T. Necrotizing fasciitis of the head and neck. Head Neck 2010;32:1592-6.
6. McAllister P, O'Neill F, Bharadwaj G, O'Regan B, Laverick S. A presentation of facial necrotizing fasciitis with orbital involvement. J Surg Case Rep 2013;2013(1).
7. Poitelea C, Wearne MJ. Periocular necrotising fasciitis--a case report. Orbit 2005;24(3):215-7.

8. Lazzeri D, Agostini T. Eyelid and periorbital necrotizing fasciitis as an early devastating complication of blepharoplasty. Plast Reconstr Surg 2010;126:1112-3.
9. Shindo ML, Nalbone VP, Dougherty WR. Necrotizing fasciitis of the face. Laryngoscope 1997;107:1071-9.