A Rare Case of Periorbital Necrotizing Fasciitis associated with Mortality following Herpes Zoster Ophthalmicus – A Case Report

Rhuen Chiou Chow¹, Chin Tuan Ong², Tengku Ain Kamalden³

ABSTRACT

Introduction: Necrotizing fasciitis following cutaneous herpes zoster is rare. Among three reported cases worldwide, one involved the head region. None had fatalities.

Case report: We report a seventy year-old Malay gentleman who presented with left painful periorbital swelling for six days, preceded by vesicular rashes. There were associated necrotic patches involving both upper and lower eyelids with foci of purulent discharge from the medial and lateral canthus. He was treated empirically with intravenous ceftriaxone, oral and topical acyclovir. Skin swab cultures yielded group B streptococcus and S.aureus, both sensitive to penicillin. Antibiotics were switched to cloxacillin and metronidazole, followed by bedside debridement of the eyelids. Despite clinical improvement, supported by normalization of biochemical markers, he succumbed to sepsis related complications.

Conclusion: Periorbital necrotizing fasciitis can be fatal. This represents the first reported mortality associated with periorbital necrotizing fasciitis secondary to cutaneous herpes zoster. Additional similar case reports would be invaluable to guide future management of this rare entity. **KEYWORDS**: herpes zoster ophthalmicus, mortality, Necrotizing fasciitis, periorbital

¹MD, ²FRCOphth, ³DPhil

UNIVERSITY OF MALAYA EYE RESEARCH CENTER & THE DEPARTMENT OF OPHTHALMOLOGY, FACULTY OF MEDICINE, UNIVERSITY OF MALAYA, MALAYSIA Corresponding author mail: <u>rhuen_chiou@yahoo.com</u>

Conflict of interest: The authors declare that they have no competing interest

INTRODUCTION

Periorbital necrotizing fasciitis (NF) is a rapidly progressive, devastating soft tissue infection associated with significant morbidity and mortality.^{1,2,3} While NF is well recognized as a serious complication of primary varicella zoster infection, its incidence following cutaneous herpes zoster infection is extremely rare.² This case represents the second case of periorbital NF arising from shingles in an immunocompetent patient. Although the patient in the previous case recovered uneventfully, our case is associated with mortality. This highlights the aggressive and destructive nature of this uncommon condition.

CASE REPORT

A seventy year-old male patient with underlying hypertension and recent cerebrovascular accident presented with severe left periorbital swelling and pain of six days duration. This was preceded by two days of vesicular rash evolving into crusting, itchy skin lesions extending from his left forehead, eyelids and cheek. He had been unwell with fever and lethargy for a week. Our patient denied history of trauma, insect bites or recent facial surgery however.

On admission, he was febrile and hypotensive, requiring fluid resuscitation. Eye assessment revealed gross left periorbital swelling and erythema with overlying excoriations and necrotic patches involving both upper and lower eyelids. Foci of purulent discharge ware present at the medial and lateral canthal regions. (Figure 1) Pupils were reactive bilaterally with no relative from afferent pupillary defect. Apart chemotic conjunctiva, the anterior segment showed no signs of inflammatory reaction. Fundal view was hazy due to significant cataracts.



Figure 1: Gross periorbital erythema with necrotic patches involving the upper and lower lids on presentation. Foci of pus discharge present at medial and lateral canthi.

2015

Laboratory parameters showed marked neutrophilic leukocytosis (22.1 $\times 10^{9}$ /L), and thrombocytosis with fibrin clot seen. Both serum urea (11.8 mmol/L) and creatinine (252 mmol/L) were elevated. Creactive protein was raised (177 mg/L) while random blood sugar was 10.0 mmol/L. Computer tomography (CT) scan of brain and orbits revealed enhancing subcutaneous edema and stranding along left scalp extending down to left periorbital region. There was an enhancing collection seen anterior to the left globe measuring 0.6x1.8cm. The left globe and intraocular structures were preserved. (Figure 2)



Figure 2: Enhancing collection anterior to left globe. Subcutaneous edema and stranding along left scalp and periorbital region seen.

Our patient received a stat dose of intravenous (IV) ceftriaxone 2g for severe sepsis with acute kidney injury, likely secondary to left periorbital necrotizing fasciitis following herpes zoster ophthalmicus. Oral acyclovir 800mg five times a day was commenced, along with topical acyclovir to his periorbital lesions.

He showed systemic improvement while on IV ceftriaxone (fever resolved 2 days later), with gradual reduction in leukocytosis from 22.1 to 7.3x10⁹ /L and normalization of renal function within five days. C-reactive protein also reduced to 77 mg/L from initial 177 mg/L by day ten. Swab culture and sensitivity yielded Streptococcus group В (penicillin sensitive) and Staphylococcus Aureus (methicillin sensitive). Blood and urine cultures were negative. Ceftriaxone was subsequently changed to IV cloxacillin 2g OD and IV metronidazole 500mg three times in day. Clinically, there was slight improvement of the periorbital swelling and erythema with less indurated areas. There was no progressive blurring of vision. Immediate debridement was delayed in view of response to medical treatment, and the areas of necrosis remained fairly static. A repeat CT orbit a week later showed reduction in left fronto-parieto-temporal previous collection and cellulitis with no new changes. The left periorbital collection was unchanged however.

Unfortunately he deteriorated acutely due to cardiogenic shock secondary to acute coronary syndrome. Bedside debridement of slough and necrotic skin over his left upper and lower eyelids was performed. There were no liquefied pus pockets. His wound was left and dressed with open Jelonet and Chloramphenicol ointment. He finally succumbed cardiorespiratory to arrest thirteen days after admission.

DISCUSSION

NF is a rare, life threatening infection of the soft tissues. Inflammation rapidly progresses from the fascia, muscles, and subcutaneous fat layers, with subsequent fulminant necrosis of overlying skin.1 It seldom affects the head and neck region (<5%), and involvement of the periorbita is even rarer, with less than 50 welldocumented cases over the past five decades.¹⁻³

Among patients developing NF, more than 50% had predisposing medical conditions, most commonly diabetes mellitus (30%).¹ More specifically, reported risk factors for periocular NF include eyelid trauma following blunt injury, insect bites, and blepharoplasty.³

NF resulting from primary varicella zoster infection in children and adults is well documented, albeit uncommon. Conversely, cutaneous herpes zoster (shingles) leading to NF is a very rare association. ⁴ To our knowledge, only three previous cases have been published to date. The first case published in 1998 involved a 76 year-old lady on immunosuppressants developing NF of her shin following disseminated herpes zoster.⁵ In 2000, NF secondary to shingles of the T4 dermatome was reported in a healthy 26 year-old lady.⁴ The most recent case in 2012 is an immunocompetent 63 year-old lady developing NF as a complication of shingles affecting her ophthalmic branch trigeminal nerve.² This is the only reported case of periorbital NF arising from herpes zoster ophthalmicus. More importantly, our case is associated with mortality, unlike antecedent reports.

Diagnosing NF is a challenge, especially in the initial stages, as it closely

mimics cellulitis or abscesses. Failure of early diagnosis occurs in over 85% of cases. Lancerotto et.al reports pain out of proportion to physical findings as the most constant initial clinical feature. Others include erythema, tenderness, swelling, and blistering.¹ Wong et.al has proposed the Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) Score System to stratify patients into low-, intermediate-, and highrisk categories early in the disease. (Table 1)⁶

Variable, Units	β	Score
C-Reactive Protein, mg/L		
<150	0	0
≥150	3.5	4
Total white cell count, per mm ³		
<15	0	0
15-25	0.5	1
>25	2.1	2
Hemoglobin, g/dL		
>13.5	0	0
11-13.5	0.6	i
<11	1.8	2
Sodium, mmol/L		
≥135	0	0
<135	1.8	2
Creatinine, umol/L		-
<141	0	0
>141	1.8	2
Glucose, mmol/L		-
<10	0	0
>10	1.2	ĩ

Table 1: The Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) score

A LRINEC score of ≥ 6 should raise the suspicion of necrotizing fasciitis, and a score of ≥ 8 is strongly predictive of this disease.

Table 1: The Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) Scoring System.

Management of periorbital NF is equally challenging. Mortality rates for NF affecting the scalp and upper face are lower (12.5%), compared to NF of the lower face $(32\%)^7$ or elsewhere (20-50%).¹⁻² Till date, early surgical debridement coupled with aggressive systemic antibiotics is the mainstay of treatment. This is followed by reconstructive surgery to achieve acceptable functional and cosmetic outcomes.¹⁻³

However, there is emerging evidence for a novel, conservative approach to NF involving the eyelids. Owing to the unique anatomical arrangement of eyelids, these cases behave differently and warrant an alternative treatment approach. The highly vascular eyelids allow better delivery of antibiotics to the infected site. In addition, natural barriers of the eye such as the orbicularis muscle can limit spread of infection towards the globe.

Since the skin of eyelids is thin and externally located, necrosis takes place early and hidden infection less likely. Patients therefore tend to present early in the disease. Luksich et.al has hence advocated intravenous antimicrobials with delayed bedside debridement after autodemarcation of necrotic edges. This protocol is suited for patients with rapid demarcation of necrotic edges, no orbital extension, minimal systemic toxicity, and close monitoring in place. Allowing autodemarcation translates into tissue conservation, and significantly less tissue is debrided compared with extensive debridement in advancing infections.⁷

In a case series of seven patients with NF involving the eyelids, five patients underwent surgical debridement of the autodemarcated necrotic areas, while two others did not require debridement at all. All subjects were treated simultaneously with broad-spectrum antibiotic therapy, and retained adequate lid function without reconstructive surgery. There were no associated mortalities.⁷ Hu et.al similarly reports an unusual case of non-progressive periorbital NF that responded well to systemic antibiotics and delayed bedside debridement.⁸

CONCLUSION

Periorbital NF, although infrequent, can be fatal. Despite recent literature supporting conservative management of NF affecting the eyelids, careful case selection is imperative. Failure of patients to respond with such protocol should alert clinicians to opt for more aggressive treatment methods. In our case, despite improvement of biochemical parameters, our patient could not avoid mortality secondary to sepsis related complications. Additional case reports of similar cases, and development of a clinical scoring system specific for periorbital NF would aid in future management of this rare entity.

CONSENT

Written informed consent was obtained from the patient, and his next of kin for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

REFERENCES

- Lancerotto L, Tocco I, Salmaso R, Vindigni V, Bassetto F, Necrotizing Fasciitis: Classification, Diagnosis, and Management. *Journal of Trauma and Acute Care*; 72(3); 560-566.
- 2. Fung V, Rajapaske Y, Longhi P, Periorbital Necrotizing Fasciitis Following Cutaneous Herpes Zoster. *Journal of Plastic, Reconstructive & Aesthetic Surgery* 2012; 65; 106-109.
- 3. Tambe K, Tripathi A, Burns J, Sampath R, Multidisciplinary Management of Preiocular Necrotizing Fasciitis: A Series of 11 patients. *Eye* 2012; 26; 463-467.
- Sewell GS, Hsu VP, Jones SR, Zoster Gangrenosum: Necrotizing Fasciitis As A Complication of Herpes Zoster. *The American Journal of Medicine* April 2000, 108; 520-521.
- 5. Jarrett P, Ha T, Oliver F, Necrotizing Fasciitis Complicating Disseminated Cutaneous Herpes Zoster. *Clinical and Experimental Dermatology* 1998, 23; 87-88.
- Wong CH, Khin LW, Heng KS, Tan KC, Low CO, The LRINEC (Laboratory Risk Indicator For Necrotizing Fasciitis) Score: A Tool For Distinguishing Necrotizing Fasciitis From Other Soft Tissue Infections. *Crit Care Med* 2004: 32; 1535-41.
- 7. Luksich JA, Holds JB, Hartstein ME, Conservative Management of Necrotizing Fasciitis of the Eyelids. *Ophthalmology* November 2002; 109(11); 2118-2122.
- 8. Hu V, Turner S, Robinson F, Non-Progressive Periorbital Necrotizing Fasciitis. *Orbit* 2008, 27; 59-62.