Polymicrobial odontogenic periorbital and orbital necrotizing fasciitis (PONF): A case report

Arman Mosenia a,b, Abtin Shahlaee a,1, Isaiah Giese c,2, Bryan J. Winn a,d,*

a Department of Ophthalmology, University of California San Francisco, 490 Illinois Street, San Francisco, CA, 94158, USA
b School of Medicine, University of California, San Francisco, 533 Parnassus Ave, San Francisco, CA, 94143, USA
c Department of Ophthalmology, California Pacific Medical Center, 711 Van Ness Avenue, San Francisco, CA, 94102, USA
d San Francisco Veterans Affairs Medical Center, 4150 Clement Street, San Francisco, CA, 94121, USA

ARTICLE INFO

Keywords:
Odontogenic
Periorbital necrotizing fasciitis
Orbital necrotizing fasciitis
Polymicrobial
Streptococcus milleri group
Staphylococcus lugdunensis
Microbiology

ABSTRACT

Purpose: To present a case of periorbital and orbital necrotizing fasciitis (PONF) from an odontogenic source with a distinct microbiologic profile and highlight the need for emergent multidisciplinary management.

Observations: A 39-year-old man presented with periorbital swelling, pain, and erythema following facial trauma. Imaging revealed peri-dental collections, accompanying maxillary sinusitis, and pre- and post-septal involvement. Immediate surgical debridement of necrotic tissue along with broad-spectrum antibiotics were pursued for management. Cultures grew multiple organisms, most notably Streptococcus milleri group and Staphylococcus lugdunensis.

Conclusions and Importance: PONF is a rare yet potentially fatal disease. Streptococcus milleri group and a fulminant course are to be suspected when the source is odontogenic. Timely multidisciplinary surgical debridement and medical management with intravenous antibiotics is critical for best outcomes.

1. Introduction

Necrotizing fasciitis (NF), characterized by necrosis of subcutaneous tissue and fascia, is a progressive and potentially fatal soft tissue infection. Periorbital/orbital NF (PONF) is rare, with an incidence of 0.24 cases per million per year.1 Risk factors include rheumatologic diseases, immunosuppression, alcoholism, and diabetes mellitus,2 and it is frequently preceded by trauma or surgery. Here, we present a unique case of odontogenic PONF following trauma with a distinct microbiological profile and highlight the importance of immediate multidisciplinary care.

2. Case report

A 39-year-old man was referred to the emergency department with progressive left-sided periorbital swelling, erythema, and pain following blunt facial trauma two days earlier. His medical history was only remarkable for heavy alcohol use. Initial ophthalmic evaluation revealed left-sided proptosis, relative afferent pupillary defect, and diffuse restriction of extraocular motility, especially in supraduction. In the affected eye, visual acuity was limited to 20/200 and intraocular pressure (IOP) was 17 mmHg. Vision and IOP were 20/25 and 8 mmHg in the fellow eye, respectively. The left lower eyelid was tense and demonstrated an area of obvious skin necrosis and purulent discharge in the fellow eye, respectively. The left lower eyelid was tense and demonstrated an area of obvious skin necrosis and purulent discharge

https://doi.org/10.1016/j.ajo.2022.101439
Received 19 July 2021; Received in revised form 30 January 2022; Accepted 15 February 2022
Available online 18 February 2022
2451-9936/Published by Elsevier Inc. This is an open access article under the CC BY license (http://creativecommons.org/licenses/by/4.0/).
consistent with necrotizing fasciitis. Concomitant maxillary antrostomy, ethmoidectomy, and extraction of the infected tooth were performed. Re-exploration and limited debridement were performed 12 hours later. Dilated fundus examination revealed ischemic retina with serous retinal detachment over the posterior pole and central folds over the macula. The area was again explored 48 hours after initial surgery without any evidence of additional necrosis. Intraoperative cultures grew moderate oronasal flora, gram positive anaerobic bacteria, numerous polymorphic

Fig. 1. A) Initial presentation demonstrating prominent periorbital swelling and purulent discharge with skin necrosis B) Status post three rounds of exploration and debridement C) After reconstruction of defects at 2 weeks with split-thickness skin graft and Frost suture tarsorrhaphy.

Fig. 2. Non-contrast CT scans at initial presentation: A) sagittal view illustrating peri-dental lucency surrounding the first upper left molar tooth (arrow). B) Maxillary sinusitis, and significant pre- and post-septal collections on the left side concerning for necrotizing fasciitis on coronal view.
bacteria from the *Streptococcus milleri* group, and few *Staphylococcus lugdunensis*. Antibiotics were narrowed to intravenous ceftriaxone and metronidazole and were continued for 7 days. Visual acuity was diminished to no light perception on the left. At 2 weeks, the patient underwent a repair of the left lower lid and cheek defects with a split thickness skin graft (Fig. 1) due to limited vascular supply in the heavily debrided area and risk of full-thickness skin graft failure. Six-months post-operatively, the patient had expectedly developed cicatricial ectropion of the lower lid with anterior lamella shortening, which was successfully repaired.

### 3. Discussion

Cases of NF originating from an odontogenic infection are infrequent and mostly reported in the context of lower head and neck involvement. PONF secondary to an odontogenic source has only been reported in three instances. These cases were triggered by tooth extraction or an isolated tooth infection. In our case, there was a tooth infection along with periocular trauma, both of which are individually associated with NF and may have had synergistic effects. Heavy alcohol use has also been previously linked to PONF and could have contributed to the risk of developing NF in this patient.

The most common organisms in PONF are mixed flora (including anaerobic bacteria), group A beta-hemolytic *Streptococcus*, *Pseudomonas aerogena*, and *Staphylococcus aureus* (including methicillin-resistant *Staphylococcus aureus*). Compared to NF elsewhere, PONF may evolve rapidly and involve adjacent cervical, thoracic, and intracranial areas. Fibrofatty tissue invasion via enzymes and toxins that are produced by these bacteria, including hyaluronidases, lipases, collagenases, and steptokinase, facilitate their rapid spread. Consequently, mortality directly correlates with time to intervention. The first major clinical decision point at presentation is to distinguish preseptal or orbital cellulitis from PONF. Calculating the Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) score can aid in this decision (Table 1).

| Patient’s Score | LRINEC Variable | Score |
|-----------------|-----------------|-------|
| +4              | C-Reactive Protein (mg/L) | 0     |
| (186 mg/L)      | ≥ 150           | 4     |
|                  | Total white cells (10³/L) | 2     |
|                  | < 15            | 0     |
| (11.2 × 10⁵/L)  | 15–25           | 1     |
|                  | ≥ 25            | 2     |
|                  | ≥ 13.5          | 0     |
| (15.6 g/dL)     | 11–13.5         | 1     |
|                  | < 11            | 2     |
|                  | Sodium (mmol/L) | 0     |
|                  | ≥ 135           | 2     |
| (132 mmol/L)    | < 135           | 0     |
|                  | Creatinine (mg/dL) | 0     |
|                  | ≥ 1.6           | 2     |
| (0.66 mg/dL)    | > 1.6           | 0     |
|                  | Glucose (mmol/L) | 1     |
|                  | ≥ 180           | 0     |
| (132 mmol/L)    | < 180           | 1     |
|                  | Total           | 6     |

|                  | 13               |

### 4. Conclusions

We present a case of PONF with a unique presentation and combination of organisms highlighting the need for emergent surgical and multidisciplinary management. *Streptococcus milleri* group is associated with odontogenic PONF. Both *S. milleri* and *S. lugdunensis* likely contributed to PONF in this case. Clinicians should anticipate a fulminant course of disease especially when the underlying infection is odontogenic and polymicrobial.

**Patient consent**

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

**Funding**

This manuscript was supported, in part, by the UCSF Vision Shared Resource Core Grant (NIH/NEI P30 EY002162) and departmental support from Research to Prevent Blindness.

**Authorship**

All authors meet the ICMJE criteria and have significantly contributed to the creation of this manuscript.

**Declaration of competing interest**

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

**Acknowledgments**

We would like to thank our colleagues in the UCSF Departments of Infectious Diseases, Oral and Maxillofacial Surgery as well as Plastic &
Reconstructive Surgery for their assistance with the management of this case.

References

1. Flavahan PW, Cauchi P, Gregory ME, Foot B, Drummond SR. Incidence of periorbital necrotising fasciitis in the UK population: a BOSU study. Br J Ophthalmol. 2014;98(9):1177–1180. https://doi.org/10.1136/bjophthalmol-2013-304735.

2. Amrith S, Houdgara Pai V, Ling WW. Periorbital necrotising fasciitis - a review. Acta Ophthalmol (Copenh). 2013;91(7):596–603. https://doi.org/10.1111/j.1755-3768.2012.20421.x.

3. Clement CI, Hasso ME. Necrotizing fasciitis of the face and orbit following complications with a tooth abscess. ANZ J Surg. 2004;74(1-2):85–87. https://doi.org/10.1111/j.1445-1433.2003.02902.x.

4. Shield DR, Servat J, Paul S, et al. Periocular necrotizing fasciitis causing blindness. JAMA Ophthalmol. 2013;131(9):1225. https://doi.org/10.1001/jamaophthalmol.2013.4816.

5. Li E, Distefano A, Solrab M. Necrotizing orbital cellulitis secondary to odontogenic Streptococcus constellatus. Ophthalmic Plast Reconstr Surg. 2018;34(5):e160–e162. https://doi.org/10.1097/IO.P.0000000000001185.

6. Wong C-H, Khin L-W, Heng K-S, Tan K-C, Law C-O. 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(7):1535–1541. https://doi.org/10.1097/01.CCM.0000129486.35458.7d.

7. Andreoni F, Zürcher C, Tarnutzer A, et al. Clindamycin affects group A Streptococcal virulence factors and improves clinical outcome. J Infect Dis. 2016; jiw229. https://doi.org/10.1093/infdis/jiw229. Published online May 30.

8. Coyle EA, Cha R, Rybak MJ. Influences of linezolid, Penicillin, and clindamycin, alone and in combination, on streptococcal pyrogenic exotoxin A release. Antimicrob Agents Chemother. 2003;47(5):1752–1755. https://doi.org/10.1128/AAC.47.5.1752-1755.2003.

9. Matthews PC, Lazarus R, Protheroe A, Milford C, Bowler ICW. Acute necrotizing sinusitis caused by Staphylococcus lugdunensis. J Clin Microbiol. 2011;49(7): 2740–2742. https://doi.org/10.1128/JCM.00722-11.

10. You YO, Kim KJ, Min BM, Chung CP. Staphylococcus lugdunensis-a potential pathogen in oral infection. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1999;88(3):297–302. https://doi.org/10.1016/s0093-9964(99)00314-6.

11. Frank KL, del Pozo JL, Patel R. From clinical microbiology to infection Pathogenesis: how daring to Be different works for Staphylococcus lugdunensis. Clin Microbiol Rev. 2008;21(1):111–133. https://doi.org/10.1128/CMR.00036-07.

12. Shah SA, Meyer DR, Wladis E, et al. Streptococcal orbital abscesses. Ophthalmology. 2012;119(2):425. https://doi.org/10.1016/j.jama.2011.09.047, 425.e3.

13. Goawalla A, Mansell N, Pearson A. Septic cavernous sinus thrombosis with bilateral secondary orbital infection. Orbit Amst Neth. 2007;26(2):113–116. https://doi.org/10.1080/01676830600567418.

14. Jiang S, Li M, Fu T, Shan F, Jiang L, Shao Z. Clinical characteristics of infections caused by Streptococcus anginosus group. Sci Rep. 2020;10(1):9032. https://doi.org/10.1038/s41598-020-65977-z.

15. Azam D, Spellberger B. Molecular pathogenicity of Streptococcus anginosus. Mol Oral Microbiol. 2014;29(4):145–155. https://doi.org/10.1111/omi.12056.

16. Sitkiewicz I. How to become a killer, or is it all accidental? Virulence strategies in oral streptococci. Mol Oral Microbiol. 2018;33(1):1–12. https://doi.org/10.1111/omi.12192.