

Necrotizing Fasciitis of the Lower Lip Originating from a Maxillary Odontogenic Infection

Ahmed Issam Abbas Eldawi^{1, *}, Safa Abdelazim Ahmed Osman²

¹Department of Oral and Maxillofacial Surgery, Khartoum Teaching Dental Hospital, Khartoum, Sudan

²Department of Oral and Maxillofacial Surgery, Soba Teaching Hospital, Khartoum, Sudan

Abstract

Background: Necrotizing Fasciitis or flesh eating bacteria is a rare, rapid and aggressive infection of soft tissues with high morbidity and mortality rates. Rarely it may arise from odontogenic origin in the head and neck. It is not easy to diagnose Necrotizing Fasciitis and it usually involves patients with chronic diseases and compromised immunity. Necrotizing Fasciitis is also polymicrobial or from a single microorganism. Therefore culture and sensitivity tests are of great value. Case report: A 54-year-old female presented with dental infection raised from decayed upper left posterior tooth causing facial swelling and sinuses discharging pus in the face. In the next few days of admission, necrosis of the labial mucosa and vermilion border of the lower lip appeared and progressed rapidly despite the intervention done. Results: The patient was treated by hospital admission, frequent radical surgical excisions, medical support, a combination of high doses of antibiotics and frequent wound dressings. The patient was accepting of the lip defect and she refused any reconstructive procedures. Conclusion: Necrotizing Fasciitis is a life threatening infection especially in diabetic patients. Early diagnosis and surgical intervention is considered cornerstone for the treatment of Necrotizing Fasciitis. Hyperbaric oxygen therapy and Intravenous immunoglobulins G also may be considered as an adjunctive therapy whenever possible.

Keywords

Facial Necrotizing Fasciitis, Necrotizing Fasciitis, Odontogenic Origin, Head and Neck, Labial, CNF, NF

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1. Introduction

Necrotizing Fasciitis (NF) is an uncommon, fatal and rapid progressing soft tissue infection with high morbidity and mortality rates, characterized by necrosis of the fascia and skin [1-11], suppuration and systemic toxicity [4, 8, 12]. These cases are commonly seen in the lower extremities [12-15], groin [6, 16] and abdominal wall [7, 16] but not frequently seen in head and neck regions [5, 12, 16-22] which account for only 5% of cases [21, 23].

Necrotizing Fasciitis (NF) is considered very rare on the face [12] and the lips [22], but if presented is mainly odontogenic in origin [6, 8, 9, 16, 19, 20, 24] and less commonly

pharyngeal [1, 9, 12, 20] or idiopathic [6, 18, 25]. Other less common causes could be trauma [6, 12, 18, 24], post-operative infection, insect bites [6, 24, 26], burns [18], etc. The rapid spread of infection is mainly due to the polymicrobial origin and it takes place through the fascial planes [1, 8, 11, 12, 17, 20] involving the muscles and subcutaneous fat [23].

Head and neck NF is usually predisposed and/or aggravated by underlining systemic illness and compromises health of the patients [1, 6, 8, 11, 14, 17, 18, 20, 22, 25-27]. Nevertheless, it may occur in otherwise healthy individuals [2, 6, 11, 18, 27, 28]. Patients may present with “systemic toxico-septic phenomena” [13], illness, fever, tachycardia

* Corresponding author

E-mail address: dreldawiomfs@gmail.com (A. I. A. Eldawi), sfaabdalaziem@yahoo.com (S. A. A. Osman)

and dehydration [1, 2, 6, 24, 25].

Usually patients have no specific signs and symptoms and it can be easily misdiagnosed in the early stages as cellulites [2, 7, 24, 26, 29], abscess [7, 8, 29] or erysipelas [2, 24], but later on, clinical features are enough for diagnosis [1]. The skin at site of involvement can be swollen [6], erythematous, tense [1-3, 25], warm and painful. There can also be signs of bullae, crepitus, skin necrosis [2, 11, 25, 26], fascial necrosis [27] and mal odor [2, 24] of the pathognomonic dishwater pus [16]. Some cases may show bone involvement. Management of these patients is based on the radical surgical debridement of the site of involvement, high doses of antibiotics and management of the underlying systemic disease [1, 2, 23, 26].

Here we report an undiagnosed diabetic patient with NF originating from upper posterior tooth and extending through the cheek toward the lower lip, and mental region.

2. A Case Report

We present a case of a 54-year-old female referred from a private hospital to the Khartoum Teaching Dental Hospital complaining of painful swelling on the left cheek of 5 weeks duration. We discovered that after 3 days of the swelling onset the patient went to a health center where they incised and drained the swelling with antibiotics prescribed for 7 days. The tooth complained about was scheduled to be extracted after a week. After two weeks of no improvement, the condition started to worsen; the pain and swelling progressively increased. The swelling rapidly descended to involve the left half of the upper lip, most of the lower lip and chin. The patient used herbal treatment on the skin over the swelling made from Fenugreek with no improvement noticed.

On initial examination of the patient she looked ill and fatigued, the erythematous swelling was located on her left cheek, lateral part of the upper lip, lower lip and chin. There was moderate trismus. Also, there were two sinuses with a purulent discharge of offensive odor, one on the lower lip and one in the mental area. The vermillion border of the lower lip on the left half was dark reddish in color, dried and the whole area was tender. Intraorally there were signs of poor oral hygiene, generalized calculus and several decayed teeth.

Her vital signs at the time of admission were as follows: blood pressure was 105/35 mmhg, pulse rate was 85 beats per minute, breathing rate 35 cycles per minute and her temperature was 39C. When thorough assessment and lab tests were done, results disclosed the undiagnosed Diabetes Mellitus. Her blood glucose was 438 mg/dl, Sodium (Na) was 115mmol/l, Hb 10.6 g/dl, TWBCs were $8.4 \times 10^3/\mu\text{l}$, Urea

and Creatinine were 38mg/dl and 0.7mg/dl respectively. Her C-reactive protein was 24mg/l while her HIV, HBV & HCV tests were negative. Blood culture done showed no growth after 3 days. Therefore, the patient was admitted to the hospital. All other investigations were within the normal limits.

From the information mentioned above, the case was diagnosed as infection of the Fascial spaces from odontogenic origin. The first attempts made for the case to manage her sepsis were IV fluids, an antipyretic and pain killers, control of her blood glucose level and IV empirical antibiotics as presurgical doses. Paracetamol 1gm/ Diclofenac sodium 75mg, Cefuroxime Sodium 750mg and Metronidazole 500mg were given to the patient. Soluble insulin was given 6 hourly after blood glucose measuring and the patient was fed through a nasogastric tube that includes dietary supplements and multivitamins. The upper left second premolar tooth was extracted and pus was drained through the present sinuses.

A CT scan was requested but the patient was unable to do it because of her socioeconomic status. A Panoramic radiograph was done to assess the teeth and bones which showed the empty socket of the left second premolar that was recently extracted.

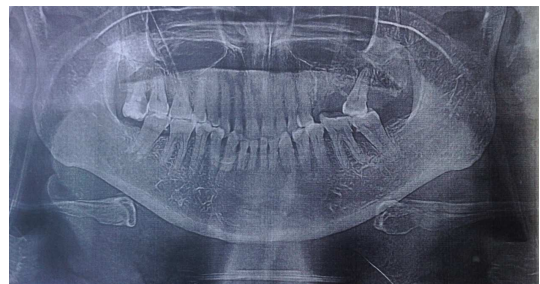


Figure 1. An OPG showing the socket of the extracted tooth.

On the second day of admission the lip became black, dried, hard and non sensitive to touch and punch. The mucosal part on the left side of the lower lip and part of the buccal mucosa were whitish in color and insensitive with areas of slough. From this clinical scenario, modification of the diagnosis of Labio-facial NF was done and the patient was sent to the septic operation room where surgical excision and debridement were done for all necrotic tissues on the left side of the lower lip vermillion border and facial mucosa involved under local anesthesia. Excised tissues and swabs from the discharge were sent for culture and sensitivity. A new incision was placed on the submental and submandibular areas to maintain drainage of the infected areas using the gravity. All wounds and the fascial spaces were explored, debrided and irrigated using NaOCl and normal saline, then all exposed wounds were packed with topical tetracycline on

a povidine moisturized gauze, and dressings were changed every 8 hours. Medications were taken and dressings changed regularly for 2 days.



Figure 2. Patient on second day of admission.

On the fourth date of admission there was no clinical improvement of the wounds, necrosis appeared again on the previously excised areas and a third sinus appeared on the labial skin just below the vermilion border. The results of the C&S were *Enterococcus* species from the excised tissues and *Staphylococcus Aureus* from the discharge which found to be resistant to penicillin.

Again the patient was sent to the septic operation room for more radical and extensive surgical excision after marking of the vital and bleeding areas using probe and a needle. Based on the C&S results, consultation with the internal physician and colleagues medications were changed. Cefuroxime was replaced by Cefazimide 6gm/day for 3 days then tapered to 4gm/day for one day and continued as 2gm/day thereafter. Gentamicin 240mg/day used for 5 days was also added to the medication list while monitoring the renal profile. Copious irrigation was done with diluted hydrogen peroxide while the dressing of the wounds was the same as previous.



Figure 3. Patient on fourth day of admission.

After two weeks of management, the patient was discharged

on oral medications and insulin. Close follow up continued for 6 months without recurrence. There was minimal wound contraction of the lip and shallowing of the facial vestibular at the left side of the lower lip. The patient was accepting of the lip defect and she refused any reconstructive procedures.



Figure 4. Patient on her regular follow up visits.

3. Discussion

The term of Necrotizing Fasciitis was introduced in 1952 by Wilson [2, 3, 6, 8, 13, 16, 20]. Other terms include; Hospital gangrene, Meleney's gangrene, Fournier's gangrene, suppurative fasciitis [6, 12, 15], necrotizing erysipelas [8], necrotizing cellulitis, necrotizing soft tissue infection, nonclostridial gas gangrene [6], malignant ulcer [24] and flesh eating bacteria [27]. Cervicofacial Necrotizing Fasciitis CNF secondary to odontogenic infection is mainly caused by the spread of microorganisms to the periapical area [1]. Researchers suggest that NF of the head and neck can be further subdivided into craniofacial and cervical NF according to the level of involvement above or below the lower border of the mandible [19, 26].

The polymicrobial nature of the odontogenic infection may initiate different diseases depending on the different synergistic combination of microorganism [1, 7, 13, 18, 30]. NF is also polymicrobial [26] or from single microorganism [25], therefore, researchers classify NF according to the microbiology into Type I: polymicrobial and Type II: monomicrobial [9, 23] with no difference in mortality rate in between. Type II NF is the most common in the head and neck [23]. A newly added third type related to marine vibrio species has been reported [9, 29] with multiorgan failure in 24 hours if left untreated [23].

Authors suggest β -haemolytic Streptococci as a frequent causative agent [12, 17]. Although most CNF were secondary to odontogenic infection, some authors stated that *Staphylococcus Aureus* (methicillin resistant or sensitive) are the most usually cultured microorganism [13], especially methicillin resistant *S. aureus* which is found in up to 40% of cultured necrotic tissues [23]. *Prevotella* and *Fusobacterium* were the least common [25] and other streptococcus strains were not cultured in most cases [13]. This difference in microbial culture may be the cause of the different clinical manifestation between CNF and odontogenic infections [13, 26]. From everything written above, the importance of culture and sensitivity tests is affirmed [26].

The mean age of patients with NF was 41 years, more aggressive in age extremities [23]. There was no significant predilection with gender [13, 23] or race [12]. CNF usually progresses rapidly in a few days, up to 14 days in some cases [13] and patients are usually febrile and predisposed by chronic illness or immunocompromisation most commonly uncontrolled diabetes mellitus [6, 8, 20, 29] alone or associated with other comorbidities [1, 13, 23, 25, 27], alcoholism, tobacco abuse, hepatic deficit [1], renal failure, malnutrition, obesity, I.V. drug abuse, HIV and malignancy [6, 23, 26]. Diabetic patients have 9 times higher mortality rate than nondiabetic patients from odontogenic NF [17, 25]. In general, mortality rate for patients with CNF reported up to 40% [3, 5, 8] and it is associated with the related medical condition more than to the wound [4].

Mandibular molars were considered the most common source of odontogenic infection triggering CNF [1, 2, 6, 13, 17, 27, 31], mainly the third molar [13]. This was not the same in this case report; the source was a maxillary posterior tooth and the infection was mainly concentrated in the lower lip and chin.

It is not easy to diagnose NF [8, 15] as it can be missed in almost 85% of cases [23, 29]. Diagnosis mainly can be taken clinically [6, 26] and from laboratory tests. Diagnosis may be confirmed histopathologically by frozen section [6, 12], but in the case presented it was not available. In challenging cases it is better to do surgical exploration [23, 29].

In the early stages, NF appears as cellulitis followed by ischemia. As the infection progress, purplish discoloration of skin and/or blisters may take place in the involved areas [5, 11, 23, 28]. A few days later gangrene develops and necrosis is apparent after a week [5, 11, 28]. These localized areas of necrosis are secondary to thrombosis of the feeding vessels [5, 28]. Local signs and symptoms such as edema, erythema, hyperesthesia, abscess and necrosis are nonspecific [1]. Lymphadenopathy is not usually common [32]. Blisters can be seen in less than half of the cases but crepitus, necrosis and anesthesia are less common [23]. These "hard signs"; blisters, crepitus, necrosis and anesthesia although more specific they are only seen in 10% - 40% of cases [23]. In this case presented blisters and crepitus weren't detected. The extent of infection is much more than identified by the skin findings [23].

A scoring system was introduced in 2004 for the diagnosis of NF by Wong et al. based on several factors called Laboratory Risk Indicator for Necrotizing Fasciitis [LRINEC] [7, 21]. It was based on WBC, hemoglobin, serum glucose levels, Na, serum creatinine, and C-reactive protein levels, and classified by according to low, intermediate and high risk [33]. Out of 13; a score of ≥ 6 raise suspicion of NF while a score of ≥ 8 is

more predictive of NF [21]. Nevertheless the outcome is unpredictable [23].

MRI and/or CT-scans are important and of great value [6-8, 27] since they show the extent of spaces involved and areas of fluid or gas aggregations [1, 5, 7, 17, 23, 26]. Unfortunately, in this case reported, due to financial issues patient cannot manage to do it.

Medical treatment alone without or with delayed surgery for the most of cases presented prior to diagnosis of CNF reflect the importance of early surgical intervention [13, 29]. Therefore, a multidisciplinary approach and urgent aggressive surgical debridement and excision are considered the key for successful treatment [1, 2, 4, 5, 7, 11, 12, 24-26] with removal of the source of infection [20] and the maintenance of patent airway [16, 25, 30].

Surgical debridement is done up to the level of vital/bleeding adjacent tissues [5, 8, 23]. The healthy tissues at the margins are recognized when skin and subcutaneous tissues can't be separated from the underlining deep fascia [4]. Incomplete radical excision of infected tissues mainly to avoid disfigurement will lead to undertreatment [5, 28] and increase mortality [23]. It is quite common for NF patients to get repeated surgeries for the debridement of tissues [5-7, 11, 14, 16, 23, 25, 26, 28, 30]. At the same time, frequent wound dressing is considered important [16, 24] because it limits the chance of getting secondary infection [6, 12]. Topical sulfadiazine [14], normal saline, betadine, H₂O₂ and povidine moisturized gauze are usually used as local dressing [12] nevertheless, the frequency of dressing is more important than the type of solution used [12].

The use of empirical antibiotics is fundamental as soon as diagnosis of NF is established [7], then after modification of these drugs is needed according to the results of the culture and sensitivity test [6, 14, 23-26]. A combination of antibiotics is usually used [5, 7, 14], such as cephalosporin, metronidazole and aminoglycoside to work against penicillin resistant staphylococci [23]. The most commonly used antibiotics are ceftriaxone, metronidazole, clindamycin, penicillin [25], meropenem, vancomycin [28], fluoroquinolone and aminoglycoside [23]. Clindamycin is suggested to be more useful in cases with toxic shock syndrome [23]. As mentioned previously antibiotics alone without surgery will not result in resolution of the infection due to the limitation of drug distribution for the infected areas caused by the angiothrombotic effect of the disease [23].

Hyperbaric oxygen therapy (HBO) is sometimes used as an adjunct therapy for CNF patients [10, 11, 18] as it hastens wound healing, resists anaerobic bacteria and supplements immunity against infections [6, 26]. Negative pressure

therapy is occasionally used in NF [7, 21, 30] as it promotes wound healing, reducing defect size and assists in wound closure [23]. Intravenous immunoglobulins G (IVIgG) also may be considered as an adjunctive therapy [5-7, 18].

Several complications may appear in patients with NF such as permanent deformity, hemorrhage, thrombosis, airway obstruction, septic shock, DIC, mediastinitis [6, 16, 19] and multi-organ failure [16, 24]. Such complications in immunocompromised patients may lead to death [16, 20]. A clinical scoring system for the risk of death was suggested by Anaya et al. [23]. It depends on age above 50 years, body temperature over 36°C, heart rate over 110 b/m, TWBC count over 40,000/mcL, creatinine above 1.5 mg/dL, and hematocrit above 50%. A previous study showed the mean number of days for hospital admission was 36 days [23], some of it in the ICU. Nevertheless in this case, the patient was admitted for 14 days.

Later, tissue reconstruction is considered when infection is clear and patient health is retained and stable [1, 3, 14, 23, 27, 30] because these infections can result in severe deformity and even challenging in some cases [12]. Reconstruction is mainly for skin and soft tissue loss as skin grafts [11, 14, 25] and less commonly local, regional and free flaps [16]. For extensive skin loss areas, usually meshed split thickness skin graft is the best choice for coverage [4].

4. Conclusion

NF is a life threatening infection especially in diabetic patients. It needs early diagnosis, urgent aggressive surgical intervention often in series, airway maintenance, medical support and I.V. antibiotics. Frequent wound dressing is considered important using topical antimicrobial agents. Skin and soft tissue loss may need to be reconstructed.

Abbreviations

NF = necrotizing fasciitis; CNF = cervicofacial necrotizing fasciitis; CT = computed tomography; MRI = magnetic resonance imaging; OPG = orthopantomogram; TWBCs = total white blood cells; NaOCl = sodium hypochlorite; C&S = culture and sensitivity; HIV = human immunodeficiency virus; HBV = hepatitis B virus; HCV = hepatitis C virus; LRINEC = laboratory risk indicator for necrotizing fasciitis; Na = sodium; ICU = intensive care unit; H₂O₂ = hydrogen peroxide; HBO = hyperbaric oxygen therapy; IVIgG = intravenous immunoglobulins G; I.V. = Intravenous

Conflict of Interest

The authors declare no conflicts of interest regarding the

publication of this paper.

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