

Odontogenic Cervical Necrotizing Fasciitis - A Case series with Review of literature

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Abstract

Cervical necrotizing fasciitis (CNF) is an uncommon and aggressive deep neck infection, is potentially life threatening causing rapid destruction of muscles, fascia and subcutaneous fat. It clinically mimics some more benign infections (cellulitis) in its early stages. Owing to its significant mortality, it is imperative to have an early and definitive diagnosis, aided by high index of clinical suspicion, laboratory indices and radiology.

Aim: Owing to its rarity, main aim of this series is to present our clinical experience of CNF aiming to set a standard protocol for prompt diagnosis and management.

Materials and Methods: A total of 5 patients presented to ENT Department with Cervical necrotizing fasciitis from December 2011 to December 2019 were reviewed retrospectively. In this series (n = 5) of odontogenic CNF, all cases were diagnosed on the basis of clinical features, Laboratory risk indicator for necrotising fasciitis (LRINEC) score and Computed Tomography (CT) scan.

Results: Majority of cases were males (4) with co-morbidities (diabetes, hepatitis, anaemia). Although mixed flora was identified on culture and sensitivity, one had Methicillin Resistant Staphylococcus aureus (MRSA) infection. LRINEC score was correlating with the severity of infection and coordinated with Laboratory risk indicator for necrotising fasciitis - Orocervical region (LRINEC-OC) score.

Conclusion: Odontogenic infections are commonly seen but their progression to cervical necrotizing fasciitis is rare with subtle clinical findings, leading to delay in the diagnosis and management. So, ancillary diagnostic indices including LRINEC and LRINEC-OC scores should be included in the protocol.

Keywords: Cervical Necrotizing Fasciitis; Necrotizing Fasciitis; Odontogenic Necrotizing Fasciitis; LRINEC; LRINEC-OC

Abbreviations

CNF: Cervical Necrotizing Fasciitis; LRINEC: Laboratory Risk Indicator for Necrotising Fasciitis; LRINEC-OC: Laboratory risk Indicator for Necrotising Fasciitis - Orocervical Region; CT: Computed Tomography; MRSA: Methicillin Resistant *Staphylococcus aureus*

Introduction

Cervical Necrotizing fasciitis (CNF) is an aggressive and life-threatening soft tissue infection also called as necrotizing cervical fasciitis or craniocervical necrotizing fasciitis. It is characterized by rapid progression with extensive necrosis and almost always accompanied by gas formation in subcutaneous tissue, fascia and deep tissues [1]. The disease is mostly polymicrobial with

odontogenic aetiology and conditions like diabetes mellitus act as predisposing factors [2]. Mostly 2 subtypes of NF are seen depending on the microbiology - the type I and type II. Type I is polymicrobial and type II consists of infections caused by a single predominant organism [3]. Type I NF infections are often diagnosed in diabetics and Type II NF in healthy individuals.

Head and neck involvement is uncommon with a high morbidity and mortality of 40 to 76% [4]. Necrosis starts in the tissues with less vascularity such as fascia and then it extends to adjacent tissues, mainly cervical fat and muscles. The spread of pathogen along the facial planes is facilitated by bacterial toxins and enzymes. Obliterative vasculitis with micro thrombosis leads to necrosis of skin along with diminished delivery of systemic treatment to the affected region [5,6].

Early diagnosis and prompt intervention is the key, but in most cases, it is not easy to clinically differentiate from other conditions like cellulitis, due to common or non-specific symptoms and also there is diagnostic delay due to delay in seeking medical health care specially in the setup of developing countries. Hence, high index of clinical suspicion with radiological confirmation is required in every case. Owing to its rarity, main aim of this series is to present our clinical experience of CNF aiming to set a standard protocol for prompt diagnosis and management. Our work in this case series (n=5) is reported in line with CARE guidelines.

Materials and Methods

A total of 5 patients presented to ENT Department with Cervical necrotizing fasciitis from December 2011 to December 2019 were reviewed retrospectively. The diagnosis was made on the basis of clinical features, diagnostic score, CT scan and surgical findings.

Complete History followed by a thorough clinical examination was done in each case with special focus to raised body temperature, dental status and condition of skin from the cheek to the precordial region. Both routine and special investigations were done. Complete blood count, C-reactive protein, Renal and liver function tests, Blood sugar, HbA1c and serum albumin were sent in all cases on the day of admission. Sample of Pus was sent for culture sensitivity.

Demographic data, comorbidities, scoring system named Laboratory risk indicator for necrotising fasciitis (LRINEC), duration of hospital stay, complications, need for tracheostomy, ICU care and mortality were evaluated [7]. In LRINEC score only biochemical parameters were taken in account (white blood cell count, haemoglobin, sodium, glucose, serum creatinine and C-reactive protein). The maximum score is 13; a score of > or = 6 should raise the suspicion of necrotizing fasciitis and a score of > or = 8 is strongly predictive of this but a score <6 is not sensitivity enough to rule out the condition. Laboratory risk indicator for necrotising fasciitis of the Oro-cervical (LRINEC-OC) score was also evaluated. LRINEC-OC score includes C-reactive protein, serum creatinine, WBC and skin flare [8]. Maximum score is 15. Score >5 raises the suspicion of necrotising fasciitis and 9-15 score has a high risk for cervical necrotising fasciitis. Radiological evaluation was done using Computed tomography (CT-scan).

Results and Discussion

Results

Demographic data, findings and clinical course of the patients with outcome are shown in the table 1 and table 2. Out of 5 patients 4 were males (range: 35-67 years) with mean age of presentation as 52 years.

Case	Age	Gender	Focus of infection	Co- morbidities	LRINEC ^d	LRINEC-OC ^e	Bacteriology
1	35	M ^a	Dental	Anaemia	4	5	Mixed flora
2	44	F ^b	Dental	-	8	9	<i>Streptococcus fecalis</i>
3	56	M ^a	Dental	DM ^c , Hepatitis	9	10	MRSA ^f
4	67	M ^a	Dental	DM ^c , Obesity	7	6	Mixed flora
5	58	M ^a	Dental	-	7	7	Mixed

Table 1: Demographic data.

a: Male; b: Female; c: Diabetes mellitus; d. Laboratory risk indicator for necrotising fasciitis; e. Laboratory risk indicator for necrotising fasciitis of the Oro-cervical region; f. Methicillin resistant *staphylococcus aureus*.

Case	Hospital stay (days)	Duration of antibiotics (days)	Complications	Tracheostomy	ICU ^b stay(days)	Outcome
1	28	25	Thrombosis of IJV ^a	-	-	Survived
2	47	45	Cardiac arrest	Done	21	Survived
3	3	3	Sepsis, mediastinitis	-		Death
4	35	30	None	-		Survived
5	24	21	None	-		Survived

Table 2: Clinical course and outcome.

a: Internal Jugular Vein; b: Intensive Care Unit.

Patients had varied clinical presentation including cervical erythema, cervical necrosis, respiratory distress and sepsis. Most common symptoms being neck pain, neck swelling and toothache. One patient had trismus grade 2 following history of mandibular molar extraction with intraoral drainage by dental surgeon. Three Patients had co-morbidities (60%) including diabetes mellitus, hepatitis and anaemia.

Odontogenic infection was the main foci of infection in all cases with history of extraction of mandibular molar in three cases. One patient presented to emergency with respiratory distress and stridor, emergency tracheostomy was done. During the procedure patient had cardiac arrest but was revived. Discharge was done after successful decannulation.

Empirical treatment was started in all the patients on the day of reporting to the hospital with broad spectrum intravenous antibiotics, which were then changed as per culture reports. Culture demonstrated mixed flora with MRSA in one case.

LRINEC Score of >6 was noted in majority (n=4) well-coordinated with LRINEC-OC score (>5 in 4 cases). After clinical as well as radiological confirmation, surgical debridement (Figure 1 and 2) was done followed by serial sterile dressings (with hydrogen peroxide and betadine) done till the sufficient granulation tissue formed. Mortality was noted in one patient due to delayed presentation.

Mean hospital stay was 27.4 days and 1 patient required ICU care. Complications occurred in 3 patients and the most dreadful complication was mediastinitis.

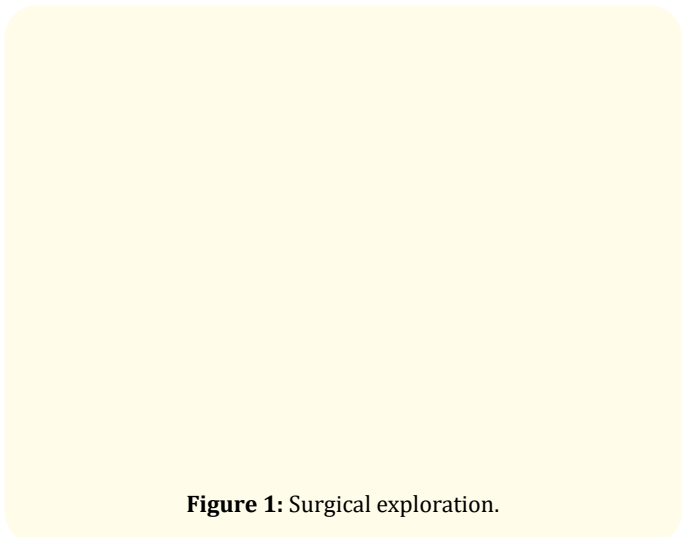


Figure 1: Surgical exploration.

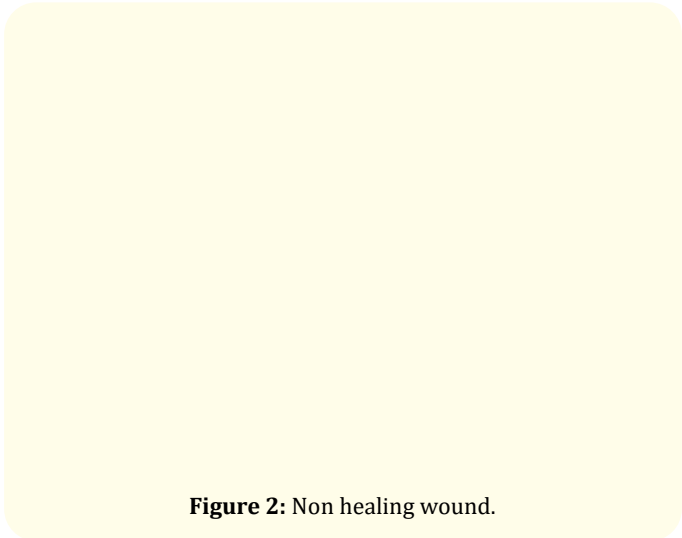


Figure 2: Non healing wound.

Mortality was 20%, cause of death was due to delayed referral with multiorgan failure and sepsis in one patient (co-morbidities: Diabetes mellitus and hepatitis).

Discussion

In the present series, the average age of presentation was 52 years consistent with the studies in the literature [9,10] but differs with the findings of others with mean age in forties [11]. This disease has a male predominance, 4 patients were males in this series. In literature male female ratio of 4:8 was also present [12]. 3 patients (60%) had coexisting co-morbidities including diabetes mellitus, hepatitis and anaemia. In this disease, there occurs liquefaction of subcutaneous tissue with disintegration of facial planes associated with venous thrombosis, inflammatory cell infiltration and endarteritis. Although most commonly affected individuals are immunocompromised with co-morbid conditions and studies have concluded diabetes as the most commonly associated co-morbidity. This illness can also affect healthy individuals without underlying systemic condition [13,14].

In our series mortality was 20%, in this patient there was delayed presentation (delay in diagnosis and referral). Also, it could be attributed to excessive pro-inflammatory cytokine response with chronic liver disease. Mao, *et al.* [15] reported a higher mortality of 40% where as a low mortality rate was reported by Lanisnik, *et al.* [16] (6%). As odontogenic infections are commonly encountered in developing nations and patients with low socioeconomic status, delayed referral and non-affordability may lead to delay in seeking treatment. One patient had trismus at the time of presentation. We found all the cases with odontogenic origin (infected mandibular molars) as the primary cause similar to other studies [13,17,18]. This is attributed to the anatomic relation of mandibular molars to the sub-mandibular space, infection spreads directly to this space with subsequent involvement of surrounding spaces.

Culture isolates in our series was polymicrobial. This is similar to literature as polymicrobial infection being commonly isolated [9,19]. MRSA was isolated in one as an unusual cause of CNF. In the literature, Zhang WJ, *et al.* reported the index case of cervical necrotizing fasciitis due to MRSA. There was rapid spread of infection despite use of antibiotics with airway obstruction and descending mediastinitis within a short period of time. Thus, MRSA in this case was no less virulent than necrotizing fasciitis caused

by any other pathogens. Hence, possibility of MRSA infection should not be underestimated and appropriate measures should be initiated if suspected [20].

Definitive diagnosis is rare solely on clinical examination findings in early stages of the disease as they mostly do not correspond to the underlying pathogenesis. To aid in early diagnosis laboratory parameters (scoring systems) have been proposed, (LRINEC) is the one widely accepted. Recently, LRINEC-OC has been proposed by Ogawa M., *et al.* [7] with sensitivity, specificity, AUC of (88.5%, 93.4%, 0.909) respectively and considered better than LRINEC score 6 [6] in patients with cervical necrotizing fasciitis. We evaluated all our cases using both the scores and to be highly indicative of the presence of CNF in majority of cases. Empirical treatment with antibiotic coverage was provided in all cases (intravenous antibiotics -ceftriaxone, amikacin, metronidazole) and following the culture reports was replaced by specific antibiotics. Radiological confirmation was done in all the cases by CT scan (due to higher sensitivity and specificity) for both diagnosis and associated complications [21]. Findings on CT scan included diffuse enhancement of fascia, muscles and subcutaneous air (Figure 3).

Figure 3: Sagittal and Axial section on CT Scan showing air in facial planes.

Hippocrates [22] first described this condition as “great falling off of the flesh, tendons and bones; and defluxion which seated in the parts was not like pus, but a sort of putrefaction.” Wilson coined the term necrotizing fasciitis in 1952 while explaining a series of streptococcal infections [23]. The involvement of cervical region is rarely seen but is mostly associated with high mortality [24].

The rarity of CNF is supported by the existing literature mostly available only in the form of case reports and case series except

only a few largest series being Lin, *et al.* [25], Mtenga AA, *et al.* [26], and Lanisnik, *et al.* [16].

Lin and co-authors evaluated 47 cases over a period of 12 years with an overall mortality rate of 25.5%. Mtenga AA, *et al.* [26] studied 151 patients over a period of 1 year with odontogenic infection. Only 42 patients met the diagnostic criteria for CNF and were included in the study. Most common co-morbidity was anaemia (11.9%) followed by diabetes.

Lanisnik, *et al.* [16] studied 34 cases of CNF over a period of 14 years and classified CNF into cervical and cranial types with reduction in mortality after aggressive surgical therapy.

Mean hospital stay in the present series was 27.4 days. Hyperbaric oxygen has been considered in the late 20th century in order to increase the bactericidal action of neutrophils, as there is low oxygen tension in the affected area. A decrease in the duration of hospital stay was noted with adjuvant use of hyperbaric oxygen in literature [13,17].

In the present series all patients underwent surgical debridement and serial dressings as the main modality of treatment. Sterile dressings were done in all the cases, till the healthy granulation tissue was encountered. Post-surgery prognosis was good. Antibiotic therapy alone is not considered best for the management of CNF, should be associated with surgical exploration. Hence, regardless of the aetiology and early antibiotic treatment, prompt diagnosis with aggressive surgical debridement is critical in the management of CNF.

Conclusion

Cervical necrotising fasciitis, a rare polymicrobial soft tissue infection of the neck is associated with high morbidity and mortality. Pre-existing co-morbid conditions like diabetes mellitus predisposes the patients to cervical necrotising fasciitis and worsens the prognosis making it imperative to aim for prompt diagnosis and aggressive management. Debridement of necrotic tissue, serial dressings and intravenous antibiotics is required. Not a common disease but clinician should be aware of the possibility of this illness. Due to delay in the diagnosis and management, ancillary diagnostic indices including LRINEC and LRINEC-OC scores should be included in the protocol.

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Ethical Approval

All procedures performed in study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Conflict of Interest

The authors declare that they have no conflict of interest.

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