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ABSTRACT

Introduction: Necrotizing fasciitis is a disfiguring condition that can be fatal. It is an aggressive infection with high morbidity and mortality. This article reviews demographics, presentation, cause, clinical findings, treatment and prognosis of cervical necrotizing fasciitis of odontogenic origin.

Materials and methods: We present 5 cases of cervical necrotizing fasciitis of dental etiology and discussed salient diagnostic features and treatment. We have reviewed the published data on cervical necrotizing fasciitis till date, with special emphasis on morbidity, mortality and treatment.

Results: With a timely diagnosis we were able to diagnose and appropriately treat these 5 patients. The most common significant medical condition in the patient history was diabetes, malnutrition. All patients were treated surgically and given broad spectrum antibiotics. All patients have survived. Of 144 patients, 27 reportedly died despite their aggressive therapy.

Conclusion: Early diagnosis and treatment of cervical necrotizing fasciitis require a high index of suspicion, immediate operative intervention, intravenous broad spectrum antibiotics and appropriate supportive care. The key to successful management and reduce the mortality is early diagnosis of the disease with prompt surgical and medical intervention. On review of literature the mortality rate was 18.75% in our study, which suggest that flesh eating disease is still a fatal disease.

Keywords: Cervical necrotizing fasciitis, Cervicofacial necrotizing fasciitis, Necrotizing fasciitis of head and neck, Necrotizing fasciitis, Soft tissue infection of head and neck, Flesh eating disease.


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INTRODUCTION

Cervical necrotizing fasciitis (CNF) is a rare, potentially devastating, severe and life-threatening, bacterial soft tissue infection that spreads rapidly along the fascial planes, causing necrosis of superficial and deep fascia, subcutaneous tissue, overlying skin and blood vessels with characteristic sparing of muscles and bone, unless they are part of original trauma or secondary to long standing untreated infection.1

Most commonly necrotizing fasciitis (NF) presents in the extremities (leg 33%, hand 7.5%)2 trunk, genitalia and perineum (20.2%).1,7 Rare in head and neck region (cervical 5.3%)1,4,6 possibly due to the great vascularity of these regions, which makes the tissue less susceptible to ischemia and infarction,7 but when occurs, dental infection is a frequent cause.4,8 Historically, NF was described in literature under various names, first described by Hippocrates in the 5th century.2,8 Different terms for NF are hospital gangrene1,9 necrotizing erysepelas, streptococcal suppurative fasciitis,1 progressive bacterial synergistic gangrene, Fournier’s gangrene and flesh eating disease.2,9 In 1952, Wilson coined the term NF.10 NF is the designation of choice, because it best describes the pathognomonic course of disease.1

We experienced 5 cases of CNF over a 7 years period. This article describes the clinical presentation, diagnosis, treatment, prognosis of our 5 cases and review of literature of previously reported cases of CNF, in addition to the role of CT, MRI and factors affecting morbidity and mortality.

CASE REPORTS

Case 1

A 35-year-old man underwent extraction of the left upper second molar, developed swelling and pain of cheek and neck after 4 days. On clinical examination, he was febrile with diffused swelling extending from left cheek to neck. Abscess was drained, tissue was removed, open dressing placed. OPG showed extraction site. Intravenous broad spectrum antibiotics started. Laboratory data indicated leukocytosis and raised ESR. On second day there was increase in the necrosis of the tissue and superficial fascia had detached (Fig. 1). Again the necrotic superficial fascia and surrounding tissues were removed completely under

Fig. 1: Clinical view of case 1 showing necrotic skin and fascia
local anesthesia, wound washed with metronidazole. After 5 days healing healthy granulation tissue was seen.

**Case 2**
A 35-year-old male patient complained of swelling and pain in the neck region since 3 days, gave history of decayed tooth since 1 year, pain since 2 months, pus extrusion from chin region. The patient was diabetic since 2 years and not on medication. Patient was disturbed and his sleep and appetite were reduced. Patient had fever, trismus and habit of chewing beetle nut. He has angular cheilitis bilaterally, raised floor of the mouth. OPG revealed multiple decayed teeth in maxillary and mandibular molar region, periapical radiolucency with 17, 38 and 46. A diffused swelling present in right and left submandibular region, crossing the midline. A sinus opening was present on submental region from where pus extrusion was seen. The overlying skin was tensed, erythematous and shiny. Nontender on palpation, soft in consistency. A provisional diagnosis of CNF was made. Patient shifted to Department of Oral Surgery, where surgical debridement was done under local anesthesia. All necrotic tissue was removed completely along with extraction of 17, 38, 46. Intravenous antibiotics, amoxicillin, cloxacillin, clindamycin and metronidazole were given. Before surgical intervention, the patient had referred to physician for treatment of diabetes.

**Case 3**
A 55-year-old male patient reported with complaint of pain and swelling on right and left side of the face with nil mouth opening. He was febrile, with a marked swelling extending from right to left pre-auricular region extending down to the neck, crossing the midline (Fig. 2). The affected area was tense, shiny. On palpation nontender, fluctuant swelling above which air crepitus was felt. OPG showed decayed 17 and 37. His WBC count, ESR were raised, hemoglobin was less. Intravenous broad spectrum antibiotics, amoxicillin, cloxacillin 250 mg 8th hourly, gentamycin 500 mg 8th hourly, diclofenac sodium 75 mg 12th hourly started, surgical debridement was done under general anesthesia (Fig. 3). CT scan showed air bubbles in the soft tissue. Stab incision and drainage was done bilaterally in submandibular region and corrugated rubber drains placed. All spaces were opened using Hilton sinus forceps. The exudate collected was thick ‘dishwater’ like, foul smelling about 30 ml on first day, 10 ml on second day. On third day skin over neck region found to be necrotized, so vigorous surgical debridement was done by giving a vertical incision on suprasternal space, one more drain placed over sternal region. Wound irrigation was done with metronidazole. On 6th day, again the wound debridement done under local anesthesia to remove the necrotic tissue, superficial fascia completely. The wound was closed by saforelle. Daily dressing was changed the patient developed breathlessness due to plural emphysema and immediately shifted to medical college, for treatment of the same. Daily wound irrigation and dressing continued. Patient recovered by 15th day of hospitalization. Later split thickness skin grafting was done.

**Case 4**
A 50-year-old male patient reported with complaint of swelling and pain on left side of the face since 5 days with nil mouth opening. Patient had fever. On inspection, a diffused swelling was extending from right to left angle of the mandible, from cheek bilaterally to the neck, crossing the midline. Overlying skin was tense, erythematous with no clear demarcation between normal and affected skin. On palpation swelling was fluctuant, air crepitus was felt. OPG showed periapical radiolucency with 37 and 38. Laboratory investigations revealed leukocytosis raised ESR, reduced hemoglobin and RBC count. Surgical debridement was
made, along with extraction of 37 and 38 teeth, all spaces were opened using sinus forcep, rubber corrugated drains were placed to keep the openings patent. Wound covered with dressing. Intravenous antibiotics, cephataxime 500 mg 8th hourly, gentamycin 80 mg 12th hourly, diclofenac sodium 75 mg 12th hourly started. On 5th day healthy granulation tissue was seen (Fig. 4) and drain was closed as pus extrusion stopped.

**Case 5**

A 55-year-old female patient reported with a complaint of pain and swelling, gave history of extraction, after which her problem started. She was anemic, febrile and restless. Her laboratory finding revealed raised ESR and WBC count, reduced hemoglobin and RBC. Microbiology revealed Gram positive bacilli. CT scan showed air bubbles and extension of lesion into mediastinum. The diagnosis of CNF with mediastinitis made. Intravenous amoxycillin, clavulanic acid, azithromycin, ofloxacin were started. The surgical intervention was done under general anesthesia; the necrotic tissue was removed completely. Wound irrigated with metronidazole and providone, closed with gauge. The features of the 5 patients are summarized in Table 1. The final diagnosis was confirmed at surgery by the presence of necrosis of subcutaneous tissue and superficial fascia.

**REVIEW OF LITERATURE**

The literature was reviewed to locate patients with cervical necrotizing fasciitis caused by odontogenic infection. A total of 144 cases of CNF of odontogenic origin reported in the literature till date in English language literature. Umeda et al reviewed and reported 125 and 9 cases respectively, of CNF of odontogenic origin. Rest of cases of CNF of odontogenic origin reported are, 4 cases by KC Ndukwe in 2002, 7 by Christopher C et al 2004, 1 by Faisal A et al in 2009, 2 by Faisal A et al in 2009, 2 by Trevor Treasure et al, 2 by Mohd Mokhtar et al 2010, and one patient by Umesh et al in 2011.

**AGE AND SEX**

Patients ages ranged from 12 to 82 years (mean, 45.2 years) and where dispersed almost equally among all generations of dentulous people. There was a slight male predominance in the patients.

**ORIGIN**

Origin of infection was mandibular teeth in 76 patients, maxillary teeth in 16 patients and origin was not described in 52 patients. In particular periapical infection of the mandibular molars, pericoronitis and impacted 3rd molars were common origin of this disease.

**ASSOCIATED DISEASES**

Eighty of the 144 patients had some associated diseases. The most frequent condition was diabetes, which was reported in 35 patients, followed by alcohol abuse in 23;
hypertension in 17; tobacco abuse in 16; corticosteroid administration and heart disease in 7 each; renal failure, drug abuse and HIV infection in 6 each; liver cirrhosis; schizophrenia in 5 each; obesity, anxiety disorder and cerebral infarction in one each. On the other hand 64 patients had no systemic disease.

TREATMENT
Most patients underwent surgical treatment and administration of intravenous antibiotics, although the details of surgical procedure, like extent of debridement, anesthesia, airway management and so forth, were not described in many reports. Thirty four patients underwent a single surgical procedure, whereas 69 patients underwent more than 2 debridement, with respect to the time before surgical intervention, 53 patients underwent surgery within 24 hours after admission, 22 patients after more than 24 hours; however, in 69 patients the timing of the surgical procedure was not described. Some patients underwent HBO and IVIG (Intravenous immunoglobulin) therapy, but the efficacy of this is not clear.

PROGNOSIS AND FACTORS AFFECTING MORTALITY
Of 144 patients, 27 (18.75%) reportedly died despite their aggressive therapy. The mortality rate of patients with associated diseases was significantly higher than that of those who did not have systemic disease. Among these diseases, the mortality rates of patients who had diabetes or who abused alcohol were significantly higher than for those, who did not have these problems. The patients who underwent surgery within 24 hours after admission showed a significantly lower mortality rate, than did those who waited more than 24 hours. Furthermore the mortality rate of those who had the mediastinitis was higher than that for patients without mediastinitis.

DISCUSSION
CNF of the head and neck is characterized by thrombosis of feeding vessels as they pass through the infected fascial planes, extensive necrosis and gas formation in the subcutaneous tissue and superficial fascia, that can lead to involvement of neck, mediastinum, chest wall and cranial base.\(^3,14\) It can be easily misdiagnosed as cellulitis,\(^11\) abscess, erysipelas\(^1,3,10,11,16\) clostridial gas gangrene, echthyma gangrenosum, pyoderma gangrenosum and wet gangrene.\(^5,16\)

NF is difficult to diagnose\(^4,8,10,13\) as in early stages of disease, the skin becomes hot, smooth, tense, shiny from the underlying edema and painful without a clear demarcation between normal and affected skin\(^1,3,4,9,10,11,13,15\) as seen in our patients. But severe pain disproportionate to local findings and associated with systemic toxic manifestations.\(^2,22\) All our patients had fever and leukocytosis, which is consistent with other studies.\(^2,3,9,11,22\) Dysphagia, odynophagia, obtundation, dehydration, trismus, paresthesia and sometimes dyspnea, anemia, hypotension, Hypocalcemia,\(^10\) tachycardia and presence of crepitations are typical signs and symptoms of CNF, affected skin may be hyperesthetic or anesthetic to touch.\(^3,22\) Patient typically presents with abnormal body temperature, airway obstruction, altered sensorial, septic shock, elevated WBC >25 mg%, leukopenia and evidence of disturbed organs functions (renal, liver, cardiovascular or respiratory).\(^9,11,17\) They also tend to be dehydrated and malnourished.\(^10\)

As the disease progresses, the pathognomonic signs of CNF appear\(^1,3,10,16,19\) ... a dusky discoloration of the skin appearing as small purplish patches with ill defined borders. Concomitantly, blisters or bullae, a few millimeters in diameter will appear in the skin of the involved area. The skin beneath the blisters becomes necrotic and blue in color.\(^1,2,10,16\) Patient shows 3 zones of demarcation: a wide peripheral zone of erythema surrounding a tender purple zone, and a center zone which becomes black and necrotic with subsequent ulceration.\(^10,16\)

Risk factors for CNF include diabetes, malnutrition, obesity, peripheral vascular disease, liver disease, immunocompromised status, hypertension, alcohol, seizure disorders, arteriosclerosis, metastatic neoplasms, chronic renal failure, polymyositis, old age, adrenal insufficiency, liver cirrhosis, drug abuse.\(^1-3,5-11,20\) In our report, the predisposing factors were diabetes (Table 1) and malnutrition, which support the findings of studies.\(^1-3,5-12,20,23\) Where as remaining did not have any such systemic disease, which indicate that CNF can occur in normal person.

In our report, the infection was caused by extraction of tooth, chronic periapical infection. These findings were consistent with that of other authors.\(^1-3,7\) The infection process may be caused by any type of trauma such as surgery, insect bite, hypodermic needle injection, intravenous needle placement, burns, cuts, abrasions, contusions, dental infections, peritonsillar abscesses, boils, simple tooth extraction, or any type of skin penetrating trauma 2 to 4 days preceding the full blown condition.\(^3,6\) Cases in which a history of trauma could not be elicited may be caused by an insult too minor to be detected, or one that was forgotten.\(^1,2,6,7\) If etiology is not known then that may be considered as idiopathic NF\(^7\) in which there is no external entry of bacteria, possibly secondary to hematogenous spread or bacterial invasion through small unnoticed breaks in the epidermis.\(^7\) CNF can also be caused by, tonsillar or pharyngeal infections, postauricular
lymphadenitis, mastoid infection,19 cervical adenitis, salivary gland infections and tumor infections.12

Plain radiographs help to detect odontogenic infection and air foci,13 as we have done in present report. But plane radiographs do not show soft tissue extension4 and deeply seated air bubbles.5,6,8,13 Computerized tomography (CT), Magnetic resonance imaging (MRI) help make an early diagnosis of necrosis. MRI can detect the extent of CNF and it can identify soft tissue edema infiltrating the fascial planes, many hours prior to local gangrenous changes.2 But MRI is not available in many places and can delay the surgical procedure.2 The role of CT scan in CNF is to help detect complications due to progressive tissue necrosis, for anatomical assessment of gas bubbles, even if they are small and few in number3,6,8,9,13 prior to and after the debridement, identify vascular thrombosis8,23 demonstrate the thickening and infiltration of subcutaneous tissues, cervical fascia, platysma, sternocleidomastoid3,12,18 fluid collection in multiple neck compartments. In our report, CT scan showed air bubbles (Fig. 5) and extension of lesion into mediastinum, parapharyngeal collection of air bubbles, indenting and displacing the pharynx to right side, spreading superiorly into left infratemporal space, laterally to involve parotid and buccal space with air foci, which is consistent with the CT findings of other authors.2,13,18,23 CT also shows a bizarre shaped hypodense areas, that does not show significant rim enhancement on contrast injection.12 This is distinguishable from an abscess, which shows peripheral rim enhancement.12 Isotope scanning, frozen section biopsy, needle biopsy or finger test are other investigatory modalities.2,9

CNF is a polymicrobial infection; frequently isolated organisms include hemolytic streptococci, hemolytic and coagulase positive staphylococci, proteus mirabilis, clostridium perfringens, bacteroids fragilis, coliforms, peptostreptococci, enterobacter and pseudomonas, hemolytic pseudomonas aeruginosa and klebsiella species. The bacterial synergism cause increase in virulence,8-10,12,13,15,17 and responsible for the fulminating course of the infections.

One of our patients was complicated by pleural emphysema, another by mediastenitis which support the findings of authors.9-11,17 The average mortality rate reported in several retrospective studies is between 20 and 40%,1,3,14,20 if mediastinum involvement occurs, the mortality rate is approximately 50%.3

Factors that contribute to high morbidity and mortality include:

- Polymicrobial infection, cancer, hypotension and WBC greater than 10 to 25%2,6,18,20 presence of comorbid factors, septic shock.8
- Delayed hospitalization, diagnosis and inappropriate treatment due to difficulty in recognizing the condition, complications
- Financial constraints, late referrals from rural health clinics and long distance to the tertiary hospitals.7

However, we found that the clinical factors affecting the mortality in 144 reviewed cases were: presence of co-morbid factors especially diabetes and malnutrition.

Of 5 patients air crepitus was palpated with only two patients which is consistent with the findings of other author.13 Umeda et al8 reported 19.2% mortality rate which is comparatively lesser than that reported by other authors.1,3,14,20 We reported 18.75% of the mortality rate, which is similar to that reported by Umeda et al.8 These facts suggests that CNF is still a potentially fatal disease.

Once CNF is recognized, treatment should be hemo-dynamic resuscitation, institution of broad spectrum antibiotics, surgical debridements, frequent monitoring, wound checks.1,6 Without aggressive medical and surgical intervention, the patient usually becomes toxic, with the potential for severe cosmetic deformity, metastatic abscess formation, multisystem organ failure and eventual death.1,6,8,11,15 Adjunctive therapy including hyperbaric oxygen (HBO) and intravenous immunoglobulins (IVIG) have been advocated.21 Further clinical trials will be needed to determine their efficacy.6,23 Under debridement of soft tissues often leads to further spread of necrotizing process to adjoining areas, requiring additional surgical intervention. Multiple surgical debridements in the operating room are usually needed. Surgical incisions are made through the discolored skin down to the fascia parallel to the cutaneous nerves and blood vessels.6 The amount of debridement can be estimated by cutting to tissue that bleeds and passing a gloved finger above the superficial fascia; when inability to pass the finger is encountered, further debridement is not necessary. Foul, thin, dishwater like murky fluid will frequently be encountered, which was present in most

Fig. 5: CT findings for case 5 showing air bubbles extending into mediastenum and displacing pharynx to right side.
patients reported\textsuperscript{1,4,6,8,10,12,15,16} as seen in our patients. Metronidazole was used to wash the wound and providone-iodine gauze packs were kept to cover the wound which are changed frequently.\textsuperscript{6,23} The split thickness skin graft is necessary to achieve cosmetic and soft tissue coverage after the healing of wound.\textsuperscript{5,23} It is important to prevent pooling of secretions in the wound that may provide a culture medium for further bacterial growth. Appropriate intravenous antibiotic coverage is imperative.\textsuperscript{2,6,23} All our 5 patients underwent surgical debridement, not only incision and drainage but also extensive debridement of the necrotic fascia, subcutaneous tissue and skin.

**CONCLUSION**

CNF should be considered in diagnosis of infections of dental origin presenting, rapidly spreading swelling, erythema and fever. CNF is associated with high rate of morbidity and mortality. Dentist may encounter patients with CNF because dental infections are the most common cause of this disease. CT scan is diagnostic, sensitive and reliable tool to detect CNF.

**REFERENCES**


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