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Odontogenic infection progressing to necrotizing fasciitis: An unusual clinical emergence

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1. Introduction

Necrosis of the lip can be observed usually in association with trauma, or a chemical hazard, infection, foreign body and adverse effect arising out due to cosmetic fillers used for the lip. The term "necrotizing fasciitis" is used to describe a severe, acute, and potentially life-threatening inflammatory condition caused by streptococcal or mixed bacterial infection and propagating continuously within soft tissues [5].

Confederate Army surgeon Joseph Jones described it as "hospital gangrene" during the Civil War, during which 46% of the 2642 soldiers afflicted died from its complications [19]. In 1920, Meleney identified 20 patients in China in whom haemolytic streptococcus was the sole organism and was called as "hemolytic streptococcal gangrene," later termed Meleney's gangrene. Wilson coined the term necrotizing fasciitis in 1952 and found no specific pathologic bacteria related to the disease [1,2].

Various terms are used for necrotizing fasciitis such as streptococcal gangrene, progressive synergistic bacterial gangrene, necrotizing erysipelas, suppurative fasciitis, acute dermal gangrene and Fournier gangrene, [3] sensational headlines, such as "Killer Bug," "Flesh-Eating Bacteria" and "Galloping Gangrene" have also been termed to it.

Early diagnosis, emergency surgical debridement, and wide-spectrum antibiotic therapy are the most effective treatment options to minimize the bacterial load and mortality rate associated with these conditions [4].

Cases of necrotizing fasciitis in the cervical region have been reported but [20] the probability of an odontogenic infection in connotation with necrosis of the lip is of an absolute surprise. Dental infections are the most common aetiology for NF of face and neck followed by trauma, peritonsillar and pharyngeal abscesses, and osteoradionecrosis. Most of the NF cases of odontogenic origin reported in the literature are of descending infection into the neck.

2. Case report

A 65-year-old woman, who's a vegetable vendor by occupation, presented to the Department Oral and Maxillofacial Surgery, Faculty of Dental Sciences, M.S.Ramaiah University of Applied Sciences with acute swelling of the right side of the face associated with severe pain, fetid odour and fever since 2 days. Patient was poorly nourished and had low BMI, she appeared toxic and restless.

She gave history of extraction of grossly decayed lower right second molar three months back in a local clinic. After extraction she developed intermittent pain in the same region. 21/2 months later pain became severe with pus exudation from the 47 socket, for which she visited a hospital where she was administered intra venous (I.V) antibiotics and analgesics (Augmentin 1.2 mg I.V). The pain did not

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subside. She continued to visit the hospital every day on out-patient basis for the I.V antibiotics. Acute swelling appeared on the lower right side of the mandible. Needle aspiration was performed extraorally, which did not reveal any exudate or pus. As the swelling persisted, she was referred to our department for further management.

Extraoral examination revealed a gross swelling extending from ala tragal line to 2 cms below the lower border of the mandible supero inferiorly. Swelling involved both the lips which appeared oedematous with shining appearance of the overlying skin. A small puncture point on the skin was present at the site of previous needle aspiration. Lower eyelid showed mild oedema. On Palpation swelling was firm. Intra oral examination revealed a diffuse swelling from midline to retromolar region. On palpation it was firm, non-fluctuant. Pus exudation seen from 47 region, which appeared as an unhealed socket associated. Additionally patient had no history any drug reaction, or insect bite or co-morbidities associated. Pain was radiating in nature and continuous which was present along the length of the mandible. CBCT was done to radiographically evaluate, which showed erosion of the bone in 47 region. Correlating with the clinical features, case was provisionally diagnosed as acute osteomyelitis of mandible. The patient was given empirical antibiotics clindamycin 600 mg I.V 8th hourly, multivitamin, NSAIDS, Tab. Sporolac with betadine mouth gargle for day 1 and day 2.

The clinical presentation and treatment instituted from day 3 is presented in a tabular form to indicate the progress to necrosis in spite of timely treatment.

Day	Clinical presentation	Procedure done
Day 3	Patient had presented with same clinical features with reduction in toxic look, fever had subsided after 2 days of medicinal therapy (Fig. 6)	Under local anaesthesia surgical exploration in the 47 region was performed intraorally, Socket was debrided tissue obtained sent for histopathological investigation, culture and antibiotic sensitivity. Surgical exploration did not yield any pus. Intraoral incisions were given in buccal vestibule 47 & 43, connected to each other to decompress the infectious/inflammatory content. Betadine soaked gauze pack was placed to maintain patency and to allow the drainage of pus or collected fluid exudate. Blood and urine sample was sent for culture.
Day 4	Reduction in size and consolidation of extraoral swelling. Small ulceration was noted on the right side corner of the lip with slight brownish purple pigmented appearance of the overlying skin. (Fig. 1)	Intra oral irrigation of the socket and site of exploration was done with chlorohexidine mouth wash. We proceeded with further investigation which included CBCT of mandible for radiographic assessment of 47 region.The analysis of different sections and 3D views revealed a bicortical bone loss irt 47 region.
Day5	Crusting of the lip with hyperpigmentation over the lateral aspect of the corner of the lip measuring around 3 cm. No intraoral exudation. Extra oral swelling was consolidated	The antibiotics iv infusion was continued and irrigation with hydrogen peroxide was carried out.
day 6	Features of necrosis (dark brown pigmentation, hard on consistency and crusting) were noticeable over the lower lip. (Fig. 2) Hyper pigmentation was prominent and similar findings were noticed in the upper lip.	The culture and sensitivity report of socket debridement which was done on 3rd day, revealed the presence of Citrobacter-freundii which was resistant to cephalosporins, ampicillin and amoxicillin with clavulanic acid. Blood culture and urineanalysis revealed no traces of infection and resulted in no organisms in the samples.
Day 7	Lower lip necrosis and crusting increased. The pattern of spread from the periphery to centre was noticed and was associated with pain. Intra orally the surgical opening had healed satisfactorily and there was no pus exudation from the socket.	In order to induce check the vascularity of the tissue, multiple punctures were done over the crusted surface on the lip but there were no signs of vascularity. A part of necrosed tissue and the adjoining slough was taken for culture and sensitivity.
Day 8	Crusting of the lips was markedly increasing and was involving lower and upper lip and was crossing the midline. Sloughing was observed in the corner of the lip associated with severe pain	I.V. Clindamycin 600 mg TID was continued. Oxum spray (superoxidised solution) dressing was advised over the crusted lips.
Day 9	Shrinking in the volume of the lower lip was apparent with necrosed tissue involving the muscle underneath the lip mucosa.	An attempt of re vascularization was done by puncture of the necrosed tissue but there was no bleeding. Oxum (super oxidized solution) dressing was placed over the lip. Culture reports showed normal oral flora with Candida Albicans. Patient was advised multivitamin injection I.V stat
Day 10	There was separation of this crusted and necrosed tissue from the underlying muscle	Under local anaesthesia the necrotic issue was detached surgically and the underlying tissue and was sutured in continuity.The defect was covered with collagen and was sutured with 4–0 Vicryl. (Fig. 3) IV Clindamycin 600 mg was replaced with Faropenem 200mg orally BID.
Day 11	Signs of healing were noticed along with softening of the crusted tissue and granulation tissue with pin point bleeding from the surface.	Same medications were continued.
Day 12	Improved signs of healing were observed.(Fig. 4)	Attempt at improving the contour was done with platelet rich fibrin matrix injection underlying the tissue into remaining lip tissue.
Day 13	Tissues healed completely, but the defect of the lower lip was present with hypopigmented scar of the necrosed tissue.	Patient was discharged.

3. Discussion

Necrotizing fasciitis (NF) is a rare soft tissue infection, primarily involving the superficial fascia and resulting in extensive undermining of the surrounding tissues. The pathological changes of NF include thrombosis of blood vessels, suppuration and necrosis of the

superficial fascia with subcutaneous fat [6]. Although many underlying disease processes predispose patients to NF, three common factors are invariably present: (a) impairment of immune system (e.g., diabetes mellitus, malignancy, alcoholism); (b) compromise of the fascial blood vessels; and (c) the presence of microorganisms that are able to proliferate within this area [7].

NF is usually spread aggressively. The plane of spread of infection will be from cuits and proceeding to subcutis usually the muscle is spared. The lip is composed of muscle orbicularis oris invested by skin, subcutaneous tissue minimal fat and salivary gland (minor) In case of the lip is more of subcute salivary glands and fat then the muscle content which is relatively less. The spread of the infection is charecterised by necrosis of subcutaneous tissue and underlying fascia, with rapid spread of infection along fascial planes.[Fig. 1].

It's a rare entity to have a necrotizing fasciitis in the lip region as it is an area of tremendous vascularity. The superior labial artery provides blood supply to the upper lip with terminal branches supplying the nasal ala and septum. The inferior labial artery provides blood supply to the lower lip and part of the superior chin. These vessels course between the mucosa and the Orbicularis Oris muscle and anastamose at the midline with branches from the contralateral side. Due to anatomical borders the infection spreads along the fascial planes. During the initial phase NF maybe mistaken for cellulitis but prompt diagnosis is warranted [8].

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Necrosis of the lips is a result of thrombosis of the vessels in between the subcutaneous tissue and the muscle where there is least resistance to the spread of infection. In 1979, Fisher described seven diagnostic parameters: Extensive fascial necrosis involving the overlying skin, systemic septic involvement with reduced mental status, lack of a primary muscle infection, missing clostridium infection, lack of vascular occlusion, and finally leukocyte infiltration with focal fascial necrosis and micro-thromboses as histological findings [4]. The early stages in NF may resemble odontogenic cellulitis or an abscess. In early signs the skin usually is tonic and red. Hyperesthesia and anaesthesia can be stated by touching which was observed from local symptoms usually are rapidly progressing, pathognomonic signs are dusky and purplish patches with ill-defined borders.

Generalised features of septic shock like somnolence and hypotension may be present early after onset of the infection. The infection primarily does not affect the musculature; muscular involvement is a sign of advanced stage of the infection associated with poorer prognosis. Mortality remains still high in NF despite use of modern powerful antimicrobial drug regimens and advances in the care of the critically ill patients [4].

Considering the results of the laboratory tests that were carried out and correlation with LRINEC and this case can be categorised under intermediate risk [9]. Therapeutical strategies in patients with necrotizing soft tissue infections are early recognition followed by a through surgical debridement eliminating entire infected tissue (cutis/subcutaneous tissue, fascial layer and if affected musculature), an organism specific antibiotic regimen is the therapy of choice but before that the fluid resuscitation, enteral feeding and intensive care treatment will overcome the symptoms of septic shock.

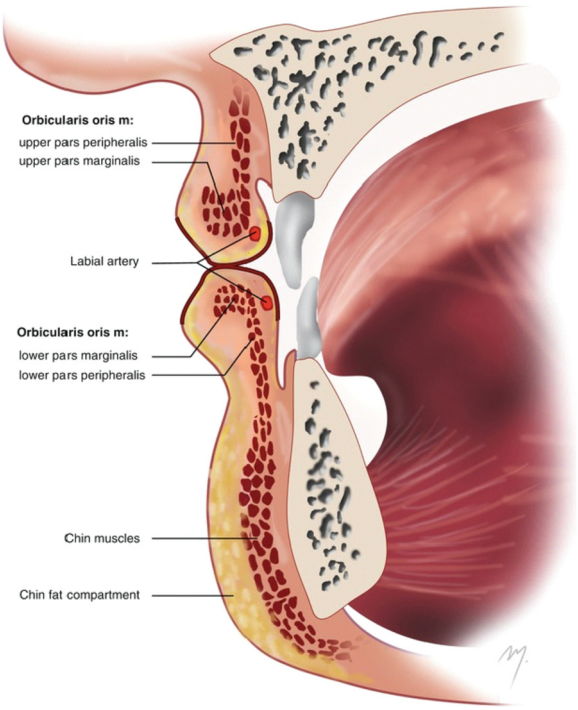

Fig. 1. Pictorial illustration of anatomy of lip.

Fig. 2. Picture depicting slight brownish purple pigmented appearance of the overlying skin.

Fig. 3. Picture depicting dark brown pigmentation, crusting of the overlying skin.

In our case, patient presented with acute signs of infection in face; there was necrosis of the overlying skin; initially there was lack of primary muscle infection but eventually the necrosis invaded the muscle, there was a missing clostridium infection and also had focal fascial necrosis and micro-thrombosis of the vessels pertaining to that area which fulfils the criteria for diagnosing as NF. There was purplish discolouration over the right side overlying the skin at corner of the mouth. However Purple, dark skin coloration and skin necrosis are signs of late stage of the disease.

We started with empirical therapy; as patient presented to us with a toxic condition. The treatment included the fluid resuscitation along with I.V antibiotics and analgesics, with multivitamin infusion. Combination therapy of Penicillin with Metronidazole or Clindamycin; third generation Cephalosporin with Metronidazole or Clindamycin; or a single antibiotic coverage with broad spectrum beta-lactam drugs such as Impenim would cover the microbial spectrum of NF. Antibiotic therapy should be continued on the basis of microbiological culture and antibiotic sensitivity test results [10,11].

Culture and sensitivity will lead us to have a clear picture of to what antibiotics to be given, surprisingly the blood and urine analysis did not reveal any traces of infection in our case. Therefore the systemic infection was negative as per blood and urine analysis report.

NF is polymicrobial and less commonly monomicrobial infection. Initially it was believed that aerobic micro-organisms, particularly group A β-hemolytic streptococci and staphylococci were considered to be the causative agents in NF. It was later demonstrated that they are strict anaerobes played a very important role in representing a mixed or synergistic infection. Micro-organisms of the Bacteroid groups; Proteus, Coli forms and Peptostreptococcus have been isolated as well as Enterobacter and Pseudomonas [12–14]. The most common organisms are group **A** beta hemolytic streptococcus and *S aureus*. Group ***C, G,*** and H streptococci, staphylococci, *Haemophilus influenzae* type B, bacteroides species, and clostridia species are also frequently isolated. Less frequently isolated bacteria are

Fig. 4. Picture depicting the necrotic issue was detached surgically and the underlying tissue and was sutured in continuity the defect was covered with collagen and was sutured.

Fig. 5. Picture depicting the healing of the tissue.

fusobacterium, actinomyces, peptostreptococcus, *P acnes, Pseudomonas aeruginosa, Proteus mirabilis*, *Escherichia coli,* enterobacter, citrobacter, and *Klebsiella pneumoniae.* [15] in present case the culture and sensitivity was done which revealed Citrobacter fruendii which was sensitive to Gentamycin and antimicrobial sensitivity was tested by modified Kirby Bauer's disc diffusion method. Other organisms which were identified as normal micro flora of oral cavity, with species of staphylococcus and streptococcus along with candida albicans.

Citrobacter Fruendii is an aerobic gram-negative bacilli. Its habitat includes the environment (soil, water, sewage), food, animals and the intestinal tracts of animals and humans [17]. As an opportunistic pathogen, C. freundii is responsible for a number of significant opportunistic infections. It is known to be the cause of a variety of nosocomial infections of the respiratory tract, urinary tract, blood and several other normally sterile sites in patients [16].

One fatal disease that C. freundii has been associated with is neonatal meningitis. Citrobacter Freundii show different antibiotic susceptibility which is why usually initial therapy is not successful. A case of necrotizing fasciitis and ostelomyelitis of the foot caused by C. freundii associated with exposure to marine animals has been reported. The isolates of C. freundii in this report states the that these are sensitive to carbapenems which have excellent activity against them [18]. In the present case carbapenem was administered which resulted in regression of the disease.

However, C. freundii has not been reported as an organism in human normal oral flora or skin, but has been reported as bacteria seen in some species of snake oral flora [21]. There have been no reports of Oral infections associated with C. Freundii resulting in necrotizing fasciitis.

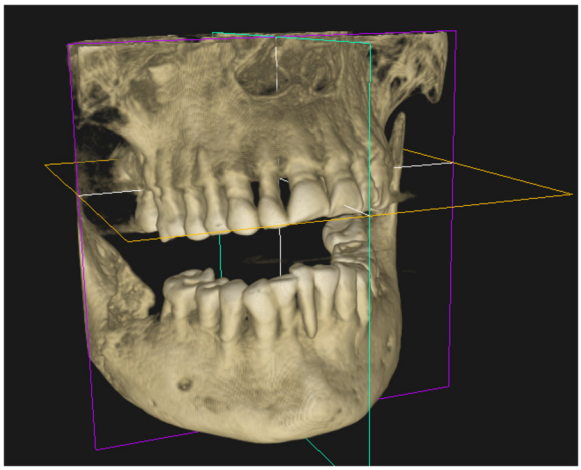

Fig. 6. Pre-op CBCT section of the jaw illustrating minimal bone loss at the site of infection.

4. Conclusion

There is an increase in occurrence of infections which are unresponsive to the commonly used empirical antibiotics due to resistance of micro-organisms. Therefore, it is judicious to closely monitor the patients and institute prompt culture and sensitivity testing, specific antibiotic to limit the spread of infection into soft tissues.

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