Necrotizing Fasciitis of the Chest Wall Caused by Infected Dentigerous Cyst: A Case Report

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Abstract Necrotizing fasciitis of the chest wall is a fairly uncommon condition. Early diagnosis and treatment is the two main factors responsible for the prognosis. Odontogenic cause of the necrotizing fasciitis of the chest wall is a rare phenomenon. It can be easily missed during the initial examination. Mortality still remains high in necrotizing fasciitis despite the use of modern powerful antibiotics and advances in the care of critically ill patients. The disease manifestation can range from a fulminant presentation to a subtle and insidious development. In this paper we are presenting a case of necrotizing fasciitis of chest wall caused by infected dentigerous cyst.

Keywords Necrotizing fasciitis · Dentigerous cyst · Odontogenic cause · Chest wall

Introduction

Necrotizing fasciitis (NF) is a rare but potentially fatal infection involving the subcutaneous tissue and fascia [1]. In the initial stages before the necrosis is seen the infection spread in the subcutaneous tissue and may appear as a routine odontogenic deep space neck abscess. Delay in diagnosis leads to increase in the area of

necrosis with a resulting increase in cosmetic deformity and life threatening complication. Death from necrotizing fasciitis can be sudden and overall mortality rate ranges from 25 to 73% in the published literature [2]. Necrotizing soft tissue infection may be rapidly fatal because of toxin induce circulatory collapse [3]. Necrotizing fasciitis can develop from dental (abscess, pulpitis, gingivitis, infected cyst etc.), sinus, peritonsillar, and salivary gland infection or infection secondary to the surgery, insect bite or trauma [3]. Dental infections are the most common etiological factor [4]. The most common organisms implicated are group A beta haemolytic streptococci, staphylococci, or obligate anaerobes [4]. Signs and symptoms of necrotizing fasciitis are high grade fever, localized swelling, erythema, induration, hypotension, tachycardia, scarletiform eruption, toxic or lethargic appearance of the patient [3]. In this case report we are presenting a rare case of NF of anterior chest wall that developed from infected dentigerous cyst.

Case Report

A 32 year old male reported to the emergency of Chadha hospital with a history of pain and swelling over the chest and neck from last 5 days. On examination swelling was present over the neck and anterior chest wall. Swelling was of approximately 7×5 cm of size in chest and 3×4 cm in neck. Swelling was tender in both sites but in chest overlying skin was necrosed. The patient was chronic alcoholic and chronic smoker. His haemoglobin was 10.5 mg/dl, total leucocytes count (TLC) revealed leucocytosis of $15,000/\text{mm}^3$ and differential leucocytes count (DLC) revealed increase in neutrophil count (80%). Blood sugar level was within normal range. Liver and renal

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function was normal. ELISA for HIV was negative. The physical examination revealed a blood pressure of 100/62 mm of Hg, temperature 37.4°C, supine heart rate 102 beats/min, respiratory rate of 17 breath/min and an oxygen saturation of 98% on room air. Immediate incision and drainage of the abscess was done at both site (Fig. 1), draining approximately 25cc of very foul smelling pus. Pus along with multiple tissue samples was sent for aerobic, anaerobic and fungal culture. Immediate complete debridement of all necrotic tissue was performed until bleeding tissue was encountered (Fig. 2). Intra-operatively patient suddenly collapsed because of cardiac arrest. However patient recovered because of immediate resuscitation procedure. Immediate inter-costal chest drain was inserted, but no purulent discharge was drained out. Beta-dine soaked gauze dressing given over the raw wound. The resected specimen grew streptococci, staphylococci and mixed anaerobes. Antibiotics cefotaxime 1 gm, gentamycin 80 mg and metronidazole 500 mg intravenously started without waiting for the culture and sensitivity test. Injection diclofenac intramuscular, serratiopeptidase and multivitamins also started orally along with parenteral fluid therapy. The wound was inspected daily to ensure tissue viability and it required further 2–3 debridement, but patient's leucocyte count was not decreasing to normal range. So after the two days detail examination of the patient was done to find out the etiological factor of the NF. Extra-orally slight swelling was present over the chin area (Fig. 3) and intra-orally a swelling was present in the buccal vestibule of the mandible extending from right mandibular first molar to left mandibular second premolar (Fig. 4). Right and left mandibular first molar was cariously exposed. Panoramic radiograph of the patient showed a unilocular radiolucent lesion present in the body of the mandible extending from right mandibular first molar to left mandibular second premolar (Fig. 5). Right and left canine was impacted. Roots of the right mandibular first molar were connected with the lesion. On aspiration pus along with the cystic fluid was aspirated. On the basis of radiographic finding and clinical examination probable diagnosis of dentigerous cyst was made. Immediately marsupialisation of the cystic lesion was done and pus along with the cystic fluid was drained out to decompress the cystic cavity. Enucleation is decided to be done later on after decrease in the size of the lesion to prevent the chances of pathological fracture. Right mandibular first molar was also extracted simultaneously. Histopathological examination confirmed the diagnosis of dentigerous cyst later on (Fig. 6). After 24 hour of the decompression of the cyst leucocytes decreases to normal range ($8,000/\text{mm}^3$). Four weeks following admission the resultant wound granulated well and ready for skin grafting which was planned to be taken from thigh area, but patient refused

Fig. 1 Showing the stab incision over the neck and necrosed skin over the chest

Fig. 2 Showing the wound after incision and drainage

further surgical treatment and allow the wound to heal by secondary intention. The patient made slow and steady progress and was discharged home in 5 week time. He continues to be followed up and is doing well.

Fig. 3 Showing extra-oral swelling over the chin

Fig. 4 Showing intraoral vestibular swelling

Fig. 5 Panoramic radiograph showing radiolucent lesion in mandible extending from right mandibular first molar to left second premolar with right and left impacted canine

Discussion

Necrotising infection of the soft tissue has been reported for centuries. However the term necrotising fasciitis was

Fig. 6 Showing histological section of dentigerous cyst (methylene blue and basic fuchsin stain, original magnification $\times 20$)

first used by Wilson in 1952 [5]. Necrotising fasciitis is most common in perineum, abdominal wall and extremities. It is less common in head and neck, especially in face and only a few cases have been reported in the chest wall [4, 6]. In most of the patient signs of inflammation at the affected site are present. Severe pain disproportionate to the local finding and in association with systemic toxicity should raise the suspicion of necrotising fasciitis. Patient presented at the advanced stage often poses diagnostic difficulties, since they may be confused, agitated, or even a reduced level of consciousness [4].

Necrotising fasciitis has been described under different names including hospital gangrene, suppurative fasciitis, meleney's gangrene, streptococcal gangrene, Fournier's gangrene, and flesh eating disease [7–9]. The variety of classification and terminology of necrotising fasciitis has been based on affected anatomy, microbial cause, and depth of infection. Terms like necrotising fasciitis, myonecrosis, and necrotising adipocytis refer to classification based on depth of infection [10]. Type 1 (polymicrobial), type 2 (monomicrobial), infection refer to classification based on microbial cause [1, 6]. Fournier's gangrene refers to infection of the perineum [10].

Organisms most commonly isolated in this form of disease are group A streptococci, staphylococci, enterococci, escherichia coli, clostridium, bacteroid, proteus, and pseudomonas [5]. The infection is polymicrobial in 70% of the cases and half of this contains mixed aerobic and anaerobic organisms. Group A streptococci has been isolated from 16 to 71% of the patients of necrotising fasciitis [10–12].

Several factors have been found to influence the final outcome of this disease. These clinical factors are old age, female sex (especially above 60 year of age), uncontrolled diabetes mellitus, anaemia, co-existing pulmonary disease, delayed referral (more than 6 days), late surgical intervention, and HIV [4, 13–15]. This patient had no comorbidities of any of the above as he was not suffering from any systemic problem and treatment was initiated within 6 days of onset of chest swelling. However patient had history of chronic alcoholism and smoking.

Imaging studies that may be useful is plain X-ray that can demonstrate subcutaneous gas, but this is a specific and not a sensitive finding. So absence of gas does not exclude necrotising fasciitis [10]. Computed tomography (CT), magnetic resonance imaging (MRI) might be useful in cases where signs are equivocal or diagnosis is in doubt. CT scan can diagnose gas in area inaccessible to palpation, identify areas where infection has spread and can also detect vascular thrombosis, erosion of vessel, or mediastinitis [4, 16]. Our patient presented late with frank area of necrosis so immediate surgical intervention was undertaken without CT scan. Immediate surgical exploration is

required if there is obvious fluctuation, necrosis of the skin, or rapidly advancing infection.

Once the diagnosis is made treatment must begin on multiple fronts. Intravenous antibiotics should be started as soon as possible and initial drug should provide effective cover against gram positive cocci, facultative anaerobes, gram negatives rods, and anaerobes [10]. In this patient we use a combination of cefotaxime, gentamycin, and metronidazole to give cover against the three principal groups of organism. Subsequent antibiotic management will be guided by sensitivity of culture taken intra-operatively. In this case culture sensitivity report showed the sensitivity of the microorganism to the already started drugs.

Aggressive surgical debridement should be done immediately and all nonviable tissue including fascia should be excised. Repeated debridement may be necessary until the infection has been controlled adequately [10]. Reconstructive surgery should be considered only once the patient has been stabilised and the infection fully eradicated [5]. No secondary reconstructive surgery has been done in this case because patient did not agree.

The role of hyperbaric oxygen in the treatment of necrotising fasciitis remains controversial. The principle behind its use is to increase the tissue oxygen tension in zone of infected hypoxic area [17].

The removal of etiological factor is also very important along with the incision and drainage and debridement of necrosed area [18]. In this patient infected dentigerous cyst was present in mandibular body region. As we decompress the cyst and drained out the pus from cystic cavity patient start recovering faster.

Conclusion

In conclusion necrotising fasciitis of the chest wall is a fairly uncommon condition and the diagnosis of its odontogenic origin may be missed at the initial presentation of the patient. The removal of etiological factor is equally important with the early surgical intervention.

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