An unusual presentation of cervical necrotizing fasciitis in an immunocompetent adult patient: a case report and literature review

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Necrotizing fasciitis is a sever and potentially fatal soft tissue infection, but its presentation in head and neck region is rare. Few reported cases of cervical necrotizing fasciitis secondary to tonsil infection were found in literature, which were proven to be dangerous life threatening condition.

In this short communication, we are presenting a 35-year-old, immune-competent Saudi female patient. She was presented to our hospital with ill looking picture of right peritonsillar abscess with mild degree of trismus. Patient was admitted, IV antibiotic was started and CT scan neck showed pus in the left tonsil parenchyma and in right peritonsillar region, with cellulitis in right parapharyngeal region with no clear collection noticed within the neck spaces. Patient was taken to OR where pus in the left tonsil surface was noted and cleaned, and incision and drainage of right peritonsillar fossae was done, pus came out and gas bubbles noted along with necrotic fleshy tissue in the lower pole of the right tonsil from which multiple biopsies were taken. Patient had postoperative bleeding after 24 hours. CT scan was repeated and patient was taken for OR for debridement of necrotic tissue from right tonsil, and right neck exploration and debridement. Necrotic tissues medial to the right submandibular gland was seen and removed. The cultured swab and aspirated pus showed coagulase-negative staphylococcus, antibiotic was changed based on sensitivity results and she responded very well. She was discharged home post operatively in stable condition and was followed up for 18 months with no history of recurrent tonsillitis.

Keywords:
cervical necrotizing fasciitis, coagulase-negative staphylococcus, peritonsilar abscess, quinsy

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Introduction

Cervical necrotizing fasciitis (CNF) is a rare life-threatening infection. It is characterize by fulminating, devastating, and rapidly progressive course, resulting in gas formation and extensive necrosis of fascia and subcutaneous tissue and later skin and muscle [1]. Despite the introduction of broad-spectrum antibiotics, mortality rate for this disease remains high [2]. It may complicate deep neck infection or may result directly from odontogenic or tonsilar infection. It is probable that cases of CNF, which arise secondary to peritonsillar abscess, have the worst prognosis, with overall mortality being higher in the group with peritonsillar abscesses (33 vs. 25%) [3]. Although necrotizing fasciitis is caused by a polymicrobial infection of aerobic, anaerobic, gram-positive, and gram-negative bacteria, CNF can be caused by coagulase-negative staphylococci [1]. Coagulase-negative staphylococcus is associated with a wide variety of infections ranging from mild skin and soft-tissue infections to serious infections include brain abscess, chronic osteomyelitis, and infective endocarditis [4]. In this report, we present a case of CNF, secondary to bilateral tonsil/peritonsillar abscess in which the isolated organism was coagulase-negative staphylococcus.

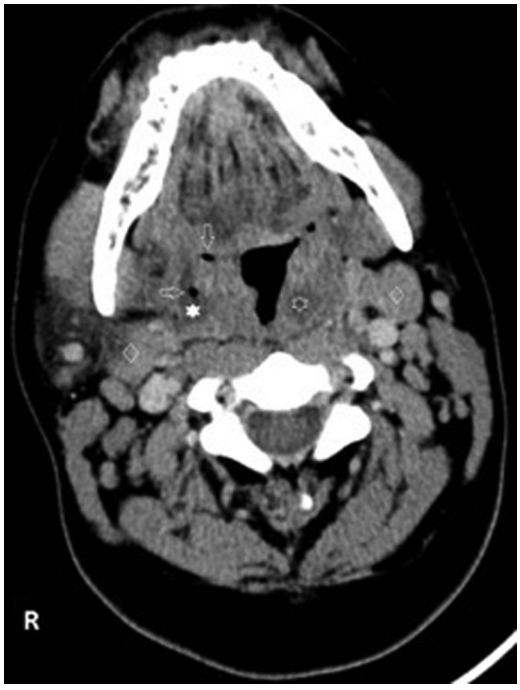
Case report

A 35-year-old Saudi woman, not known to have any medical illness, presented to our Emergency Room with complaints of sore throat, odynophagia, dysphagia, right otalgia, muffled voice, fever, malaise, and reduced oral intake of 4 days duration. She failed to respond to oral co-Amoxicillin-Clavulanic acid and Diclofenac tablets used for 3 days. There was no history of previous similar attacks nor recurrent tonsillitis, and no history of travel abroad or contact with TB-infected person. There is a positive family history of peritonsillar

abscess in brother and sister which required incision and drainage followed by elective tonsillectomy over the past 6 months. Patient works in clothes-making factory and is nonsmoker.

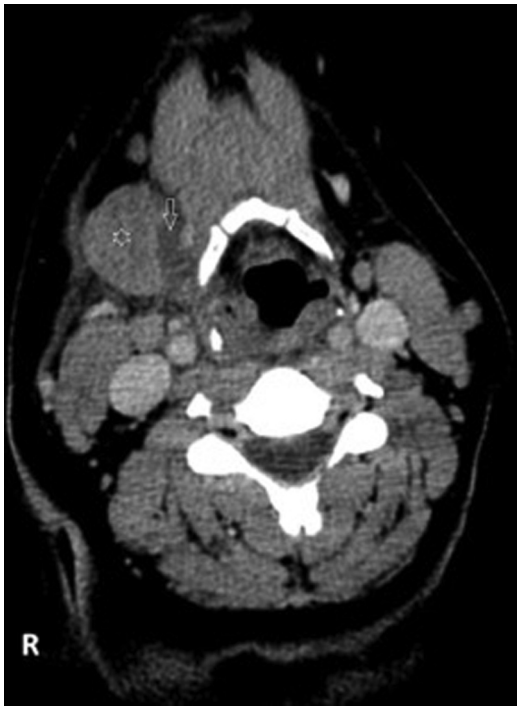
On examination, patient was obese, looking unwell, and dehydrated. There was bilateral submandibular fullness, more on the right side. The right submandibular region was tender, diffuse, without clear margins, and firm in consistency. On bimanual palpation, the right submandibular gland was enlarged, with no pus expressed from any of the salivary major gland ducts. Oral cavity examination showed grade II trismus; both tonsils were congested, large, and covered with slough; the right peritonsillar area was congested and bulged; and the uvula was pushed to the left side. Patient laboratory investigations showed leukocytosis with white blood cell (WBC) of 15 10^9/l, and erythrocyte sedimentation rate (ESR) was 125 mm/h. Trial of right tonsilar abscess aspiration was done but was not successful, as the patient was not cooperative owing to severe pain. So the patient was admitted with impression of right peritonsillar abscess. The patient was started with intravenous cefuroxime and metronidazole. On the next day, computed tomography (CT) scan of neck was advised because of persistence of severe pain that was not relieved by intravenous acetaminophen. The CT scan showed pus

Figure 1



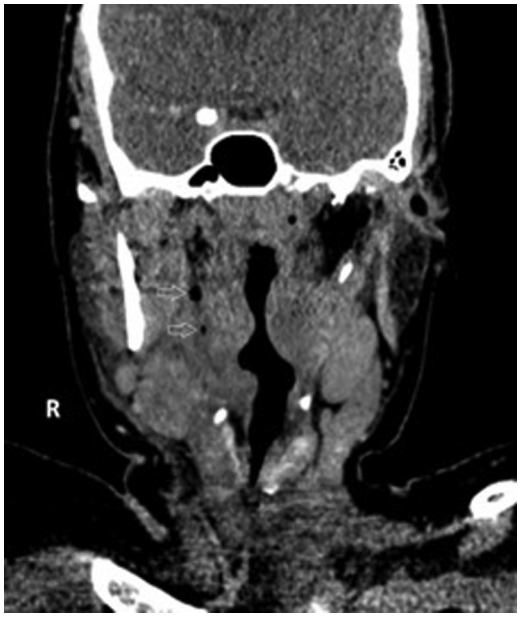
Contrast-enhanced axial computed tomographic image of the neck before first surgery: hollow arrows indicate air bubbles within the right parapharyngeal space; solid star indicates hypodense area within right peritonsilar space extending to right parapharyngeal space and obliterated fat planes and inflammatory changes surrounding it; hollow star indicates hypodense collection within the parenchyma of enlarged left tonsil; hollow diamonds indicate enlarged bilateral level IIA lymph nodes.

Figure 2



Contrast-enhanced axial computed tomographic image of the neck before first surgery: hollow arrow indicates nonenhancing low attenuation area medial to the right submandibular gland which was found to be necrotic muscle tissue during neck exploration surgery; hollow star submandibular gland.

Figure 3



Contrast-enhanced axial computed tomographic image of the neck before first surgery: hollow arrows indicate air bubbles within the right paraphygeal space.

in left tonsil and right peritonsillar area, besides cellulitis and gas in right parapharyngeal region; the right submandibular gland was enlarged with low attenuation signal medial to it. Submandibular and other cervical lymph nodes were enlarged bilaterally. There was also right maxillary sinusitis (Figs 1–3). The patient was taken to the operation room on the same day, and abscesses from right peritonsillar area and left tonsil were evacuated. The pus in right peritonsillar area was mixed with blood and air bubbles. The right tonsil looked unhealthy, hence biopsy was taken. Pus was sent for culture and sensitivity, and the results came as no beta-hemolytic streptococcus was isolated. Approximately 25 h later, patient developed severe bleeding from right tonsilar area. Therefore, she was taken to the operating room. The right tonsil was necrotic and friable, and the friable tissue was removed. The whole tonsillar fossa was oozing, so was packed with surgicel. The patient was kept intubated and admitted in ICU. Imipenem and metronidazole intravenous were started after infectious diseases consultation. WBC and ESR came down to 11.5×10^9/l and 118 mm/h, respectively. HIV Ag/Ab was negative, CMV IgM negative/IgG positive, EBV IgM negative/IgG positive, anti-HCV negative, and HBs Ag negative. Repeat CT scan showed cellulitis in the right peritonsillar and parapharyngeal areas with no evidence of abscess. The right submandibular gland was enlarged with clear and more demarcated area of low attenuation signal medial to right submandibular gland. The patient was taken to the operation theater for the third time. The peritonsillar area was opened through previous incision, and there was necrotic tissue all around the peritonsillar bed. Meticulous debridement was done. There was no active bleeding. Submandibular region was exposed through external neck incision. Necrotic muscle and fascia tissues were found medial to submandibular gland and communicating with peritonsillar fossa. Debridement was done, and corrugated drain was placed. Tissue was sent for histopathology examination and culture sensitivity, AFB stain, and TB culture. Histopathology examination revealed necrotic tissue. Tissue culture revealed coagulase-negative staphylococcus. Antibiotics were changed according to an infection disease consultant to tazobactam/piperacillin, metronidazole, and caspofungin intravenous, and the patient responded very well. On the following day, there was no drainage, so the drain was removed. On the second postoperative day, WBC became normal and ESR decreased to 55 mm/h. The patient improved significantly. Subsequently patient was extubated successfully and started on oral feeding. The peritonsillar fossa was covered with healthy slough. The external wound was healthy. On the sixth postoperative day, sutures were removed and patient was discharged on oral coamoxiclav and metronidazole. Follow-up was done in 1-week time in the clinic. The patient was healthy, with good oral intake, and the left tonsil and right peritonsillar areas were healthy.

Discussion

CNF is a rapidly spreading and potentially lethal infection characterized by diffuse necrosis of fasciae and subcutaneous tissues. Patients with peritonsillar abscess as an initial infection appear to have a poorer prognosis and have a high rate of complication and fatal outcome compared with patients with other sources of this disease [3]. Joseph Jones, an American army surgeon, described this entity in 1871 during the civil war and named it 'hospital gangrene'. In 1924, Melany reviewed 20 cases of 'streptococcal gangrene.' He was the one to note that subcutaneous necrosis is the hallmark of necrotizing fasciitis. The term 'necrotizing fasciitis' was coined by Wilson in 1952 and has gained wide acceptance [5]. It occurs in all age groups but most patients are below 40 years of age. There is no sex or race predilection [6]. Necrotizing fasciitis is classified as type I (polymicrobial), or type II (monomicrobial). Most cases of necrotizing fasciitis are type I, with 25–45% of cases being type II [7]. Fihman and colleagues, reviewed 152 patients with CNF, and found *Streptococcus milleri* group and *Prevotella* species to be the predominant pathogens, frequently copathogens, mostly of dental origin. Thus, a combined aerobe–anaerobe infection may have a synergistic effect, which allows the infection to spread in cervical tissues [8].

Coagulase-negative staphylococci have been regarded as apathogenic for long time, but their important role as pathogens and their increasing incidence have been recognized and studied in recent years. Coagulase-negative staphylococci are by far the most common cause of bacteremia related to indwelling devices. Brook and Frazier, found that isolated *Staphylococcus epidermidis* was isolated in 5 of 81 patients with necrotizing fasciitis [9–11]. Coagulase-negative staphylococci have become increasingly resistant to multiple antibiotics [12]. Most important virulence factors in the pathogenesis of foreign-body-associated infections is the ability of these bacteria to colonize the polymer surface by the formation of a thick, multilayered biofilm [13]. The diagnosis depends mainly on clinical features and a high index

of suspicion because the clinical features may be innocuous at the early stage [14]. Whitesides *et al.* [15] in a review of 12 cases of CNF found that the most common significant medical conditions in the patient's history were diabetes, hypertension, obesity, and substance abuse.

Umeda and colleagues, described the condition of this rare disease, and their objective was to find factors affecting the mortality. In his study, 24 of the 125 reviewed patients died despite receiving the appropriate therapy, factors affecting the mortality were present of associated diseases such as diabetes and alcohol abuse and delay of surgery, in addition to the presence of complications such as mediastinitis [2]. CNF that developed after peritonsillar abscesses was associated with higher mortality than that developing predominantly after odontogenic infection (33 vs. 25%) [3]. Maria and Rajnikanth [6], used markedly raised CRP as strong suspicion of necrotizing fasciitis in early stage. In our case, ESR was markedly raised on presentation, which decreased progressively with resolution of disease.

Soft-tissue radiography of the neck is a useful initial investigation in less suspicious cases, and it can detect air in the soft tissues [16]. The most common CT findings in CNF are the thickening and infiltration of subcutaneous tissues, fluid collection in multiple neck compartments, and diffuse enhancement and thickening of the cervical fascia, platysma, and sternocleidomastoid and strap muscles. Inconsistent features include gas collection in the soft tissues [17,18]. In our case, the presence of gas bubbles in CT allowed for early surgical intervention. MRI differentiates between necrotizing and non-necrotizing fasciitis [19]. MRI documented fascial inflammation, characterized by low intensity on T1-weighted images and high intensity on T2-weighted images. Absence of gadolinium contrast enhancement on T1-weighted images reliably detected fascial necrosis.

Appropriate immediate surgery and antimicrobials plus intravenous IgG therapy may be useful in the treatment of all forms of group A beta-hemolytic streptococcal-related necrotizing fasciitis (type II), especially when they are associated with toxic shock syndrome. This early combined therapy may be crucial for survival [3]. In addition to standard treatment, Safak *et al.* [20] advised hot tonsillectomy. Whitesides *et al.* [15] advised that early surgical intervention in addition to the use of hyperbaric oxygen decreases morbidity and improves the clinical outcome. Nanda *et al.* [21] used amniotic membrane effectively as a dressing material in necrotizing cervical fasciitis, which led to a significant relief of pain.

Conclusion

For an ENT surgeon, during practice, one is likely to encounter the devastating but fortunately uncommon condition of CNF. Early diagnosis, early surgical intervention, and aggressive antibiotic therapy are significant factors in reducing mortality and morbidity in CNF especially of peritonsillar origin.

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Conflicts of interest

There are no conflicts of interest.

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